The Journal of Thoracic Surgery

VOL. 23 MAY, 1952 No. 5

Original Communications

AN ANATOMIC SURVEY OF ANOMALOUS PULMONARY VEINS: THEIR CLINICAL SIGNIFICANCE

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IT IS only in recent years that attention has been focused on the anatomic variations and anomalies of certain thoracic structures. This increased interest in the variational anatomy of the thorax is but a sequel to the great advances that have been made in the field of thoracic surgery. Thus, of the total number of anomalous pulmonary veins found recorded in the literature, fully 43 per cent have been recorded within the last twenty years. In 1942, Brody[5] presented an excellent review of the literature on the abnormal connections between the pulmonic and systemic venous systems. Beginning his review with the first case reported by Winslow[22] in 1739, he collected from the literature a total of 106 cases. In 1944, Hughes and Rumore[14] cited seven additional cases which were not listed by Brody in his survey. In our own survey, we found nine other cases reported in the literature prior to 1942 and twenty-two cases which have been reported since then. Adding all cases reported in the literature to the three cases presented in this paper brings the total number of anomalous pulmonary-systemic connections thus far recorded to 147 cases.

In view of the existence of other anatomic variations, it is quite obvious that the incidence of anomalous pulmonary veins is actually higher than that which is suggested by the relatively few cases (147) reported over a period of two hundred years. Undoubtedly many additional cases have been observed during the course of two centuries, but because of lack of interest were not reported. The significance of aberrant pulmonary-systemic connections has rarely been emphasized in the past. Variations in the number of pulmonary veins occurred with such frequency that they did not merit mention in the literature, except when they were accompanied by an anomalous drainage of one or more of the veins. As far as we could ascertain, a statistical analysis of the variations of the pulmonary veins in a large series of cases has thus far not

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Received for publication Feb. 15, 1951.

been made. Therefore, one cannot come to a definite conclusion as to their incidence. The purpose of this paper is to present our observations in the dissection of 251 adult cadavers, to correlate these findings with pertinent data attainable from the literature, and briefly to review the clinical significance of anatomic variations of the pulmonary veins.

Poirier and Charpy[19] classified the variations of the pulmonary veins into two types: (1) abnormal number of pulmonary veins, and (2) abnormal drainage of the pulmonary veins, i.e., abnormal connections between the pulmonic and the systemic venous systems. The latter were subdivided into (a) complete drainage, and (b) incomplete drainage of the pulmonary circuit. In this paper Charpy's classification is retained but is modified as follows:

I. Abnormal number of pulmonary veins
  a. Decrease in number
    1. Normal drainage into left side of the heart
    2. Abnormal drainage into right side of the heart
  b. Increase in number
    1. Normal drainage into left side of the heart
    2. Abnormal drainage into the right side of the heart
    3. Mixed drainage
      a. Into left side of the heart
      b. Into right side of the heart
II. Abnormal drainage of the pulmonary veins
  a. Complete drainage of the pulmonary circuit into the right side of the heart
  b. Incomplete drainage of the pulmonary circuit into the right side of the heart

ABNORMAL NUMBER OF PULMONARY VEINS

Only a few statistical reports on the variations of the number of pulmonary veins can be found in the literature. In textbooks of anatomy, all authors describe two left and two right pulmonary veins emptying into the left atrium. Many variations are possible, and undoubtedly occur more frequently than is generally known.

The most common variation observed is the presence of a common pulmonary vein on either the right or left side (Fig. 1). Adachi,[1] in an examination of ninety-one cadavers, encountered eleven cases (12.1 per cent) having a common pulmonary vein; whereas, Healey and Gibbon[13] found fifty-two cases in 185 adult cadavers. Since publication of the latter report we have found eight additional cases in sixty-six bodies. Adding the cited figure given by Healey and Gibbon gives a total of sixty cases in 251 specimens (23.9 per cent) in which a common pulmonary vein was present. The three cases with a common vessel as described by Brantigan[4] do not lend themselves to a statistical study, for they were observed in an unspecified number of operative procedures. In each of the above described specimens the common pulmonary vein drained into the left atrium. However, there have been thirty cases reported in which a common pulmonary vein emerging from either lung drained into the right side of the heart, either directly or indirectly.

A common pulmonary vein may then be present on either the right or left side, and may drain into either the right or left atrium. The earlier writers such as Poirier and Charpy[19] state that such a common vein is not at all uncommon on the left side. Our dissections confirm this statement, for in 251

specimens we found fifty-three such vessels on the left side and only seven on the right side. The three cases mentioned by Brantigan occurred likewise on the left side. However, of the eleven common pulmonary veins described by Adachi, six were on the right side and five were on the left side. Of the thirty cases surveyed in the literature, each with an abnormal site of drainage, twenty were on the right side and ten on the left.

The next most common variation of the pulmonary veins is an increase in their number. There may be three veins from either lung entering the left atrium, but again few such cases have been reported. In our dissections this variation occurred more frequently on the right side, a separate vein being present for each lobe (Fig. 2). The additional vessel when present is known as the middle pulmonary vein. Adachi found three such vessels on the right

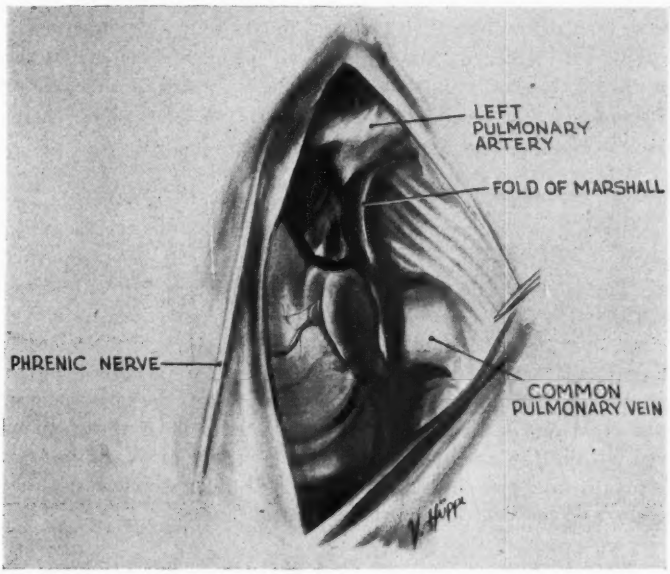

Fig. 1.—Left common pulmonary vein. (Reproduced from J. THORACIC SURG. 19: 864-874, 1950.)

side in 150 dissections (2.0 per cent). We have seen four right middle pulmonary veins in 251 bodies examined, an incidence of 1.6 per cent. Brantigan described two cases observed at operation in which three pulmonary veins were present on the right side and emptied into the left atrium. Four other cases have been reported in the literature in which three veins were present on the right side, but in each of these specimens at least one of the veins drained aberrantly. As regards the left side, Adachi encountered three separate pulmonary veins on two occasions while examining eighty-nine bodies (2.2 per cent). A specimen with three left pulmonary veins entering the left atrium was not observed in our dissections.

When more than three veins are present on one side, one or more of them, in most instances, exhibit an anomalous drainage. Such cases are relatively rare. Atkinson and associates[3] described a case in which four right pulmonary veins were present, two of which drained into the superior vena cava. Didion[10] encountered a case in which four veins were present on the right side, but all of these entered the right atrium. Two cases have been reported in the literature in which five veins drained the right lung, and in both of these cases four of the veins drained into an abnormal site. Hyrtl[16] described a case in which the left upper lobe was drained by six veins, each vein emptying into a persistent left superior vena cava.

The variations in the number of pulmonary veins are easily explained on the basis of embryologic development. Primitively, a single pulmonary vein drains both lungs. With the growth of the left atrium it becomes absorbed into the latter; so that at first one, then two, and finally four pulmonary veins empty into the left side of the heart. Should less than the normal amount of absorption take place, then a common pulmonary vein will be formed on one side or on the other. If more than the usual amount of absorption takes place, there will be an increase in the number of pulmonary veins.

ABNORMAL DRAINAGE OF THE PULMONARY VEINS

The entire pulmonary circuit may drain into the right side of the heart, either directly or indirectly. Sixty-one such cases have been reported in the literature. The most frequent sites of drainage of these anomalous vessels are shown in Table I. The average age of all the reported cases is 1.8 years; however, four of these survived beyond the age of 10 years, the oldest being 27 years. Excluding these four cases, the average age drops abruptly to 5.8 months. Survival in these cases is only possible if some form of communication between the right and left sides of the heart persists. Of the sixty-one cases, eleven lived beyond the age of 1 year. In the eleven cases, six had a patency of the foramen ovale, two had a patency of the ductus arteriosus, two had a single atrium, and one had an interatrial septal defect.

TABLE I. SITES OF DRAINAGE OF ANOMALOUS PULMONARY VEINS

COMPLETE DRAINAGE (61 CASES)	NO.	PER CENT	INCOMPLETE DRAINAGE (86 CASES)	NO.	PER CENT
Superior vena cava	10	15.9	Superior vena cava	33	35.8
Coronary sinus	10	15.9	Left innominate vein	19	20.6
Right atrium	10	15.9	Right atrium	18	19.6
Portal vein	9	14.3	Left superior vena cava	7	7.6
Left innominate vein	8	12.7	Inferior vena cava	4	4.3
Left superior vena cava	7	11.1	Azygos vein	3	3.3
Ductus venosus	3	4.7	Coronary sinus	2	2.2
Right innominate vein	2	3.1	Single atrium	2	2.2
Single atrium	1	1.6	Left subclavian vein	1	1.1
Left subclavian vein	1	1.6	Portal vein	1	1.1
Azygos vein	1	1.6	Left ventricle	1	1.1
Sinus venosus	1	1.6	Thoracic duct	1	1.1
Total	63*	100.0	Total	92†	100.0

*Two cases had two different sites of drainage.
†Six cases had two different sites of drainage.

There may be an incomplete drainage of the pulmonary circuit into the systemic venous system, and of this type eighty-six cases have been reported, including the three reported in this paper. These cases differed both in regard to the portion of the lung drained, and to the site of entrance of the anomalous vessel. In thirty-three of the eighty-six cases complete drainage of one lung was present (the right lung in twenty-two and the left lung in eleven instances). Anomalous drainage of a portion of one lung has been reported in regard to all lobes except the left lower lobe. The most frequent lobes involved were the right upper lobe (thiry-three cases) and the left upper lobe (twenty-two cases). The most frequent sites of drainage were into the superior vena cava (thirty-three cases), left innominate vein (nineteen cases), and the right atrium (eighteen cases) (Table I). Of the seventy cases in which the age was recorded, 77 per cent had reached maturity.

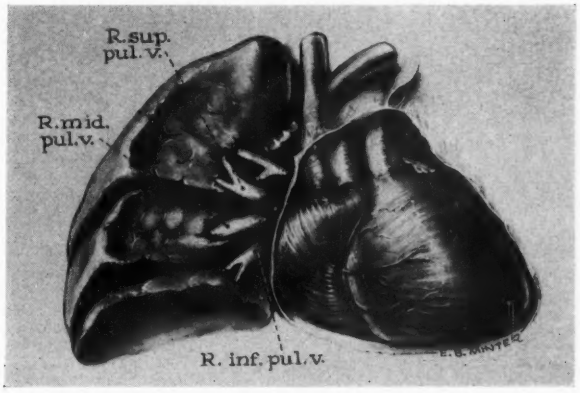

Fig. 2.—Three right pulmonary veins.

TABLE II. INCIDENCE OF ANOMALOUS PULMONARY-SYSTEMIC CONNECTIONS

AUTHOR	TOTAL BODIES (NUMBER)	ANOMALOUS VESSELS (NUMBER)	PER CENT
Adachi	270	2	0.7
Hughes & Rumore	280	2	0.7
Healey & Gibbon	251	1	0.4
Total	801	5	0.62

Adachi encountered two cases with a partial drainage of the pulmonary circuit into the systemic system in 270 cadavers. Hughes and Rumore described two cases in 280 dissections, an incidence of 0.7 per cent in both series (Table II). In our studies of 251 bodies we found one instance (0.4 per cent) of an anomalous drainage of the pulmonary veins. Since then we have observed two additional cases. A report on the three cases observed may be briefly outlined as follows:

CASE 1.—*The anomalous pulmonary drainage occurred in a white man 65 years of age whose clinical history was unobtainable and the cause of death unknown. Upon opening the thorax, a large vein was seen ascending in the superior mediastinum. This vein received tributaries from all segments of the left upper lobe including the lingular division. It ascended over the anterior aspect of the left pulmonary artery, lying parallel with the ductus arteriosus; thence it ran over the anterior surface of the aortic arch and terminated by emptying into the left innominate vein 3.5 cm. from its entrance into the superior vena cava. As a trunk, the anomalous vein measured 3.2 cm. in circumference (Fig. 3).

The left inferior pulmonary vein which drained the lower lobe emptied normally into the left atrium. Aside from the presence of a patent ductus arteriosus which measured 1.4 cm. in circumference, no other congenital abnormalities of the heart could be distinguished.

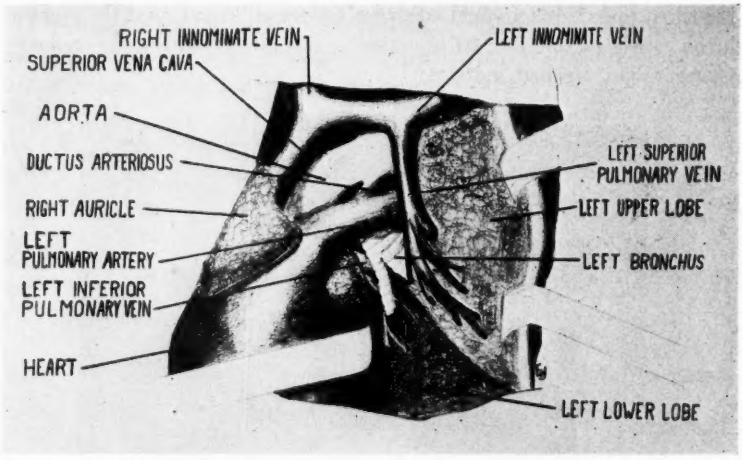

Fig. 3.—Anomalous vessel found in a dissected specimen. Left superior pulmonary vein drains into the left innominate vein. The left inferior pulmonary vein follows its normal path of drainage into the left atrium. (Reproduced from J. THORACIC SURG. 19: 864-874, 1950.)

CASE 2.—The anomalous vessel in this case was found in a 40-year-old white man. It drained the entire left upper lobe and coursed cephalad anterior to the bronchus, left pulmonary artery, and arch of the aorta. It terminated in the left innominate vein 5 cm. from the point where the latter joined the superior vena cava, and measured 4.8 cm. in circumference (Fig. 4).

The left inferior pulmonary vein was present and drained into the left atrium. The ductus arteriosus and foramen ovale were both completely sealed off.

CASE 3.—The following case of an anomalous drainage was seen in an adult male cadaver. The left upper lobe was drained by a left superior pulmonary vein, but instead of emptying into its normal site this vessel took an aberrant course, ascending into the superior mediastinum. In this part of its course it was joined by two vessels; (a) the superior intercostal vein of the left side, and (b) a vein which coursed anterior to the pulmonary artery and extended caudad into the vestigial fold of Marshall. This latter vein, however, was not continuous below with the oblique vein of the left atrium, and therefore, was not a complete persistence of the left superior vena cava. The three vessels observed, namely the left superior intercostal, the remnant of the left superior vena cava, and the left superior pulmonary vein joined to form a single trunk which entered the left innominate vein (Fig. 5).

*This case was briefly mentioned by Healey and Gibbon in J. THORACIC SURG. 19: 864, 1950.

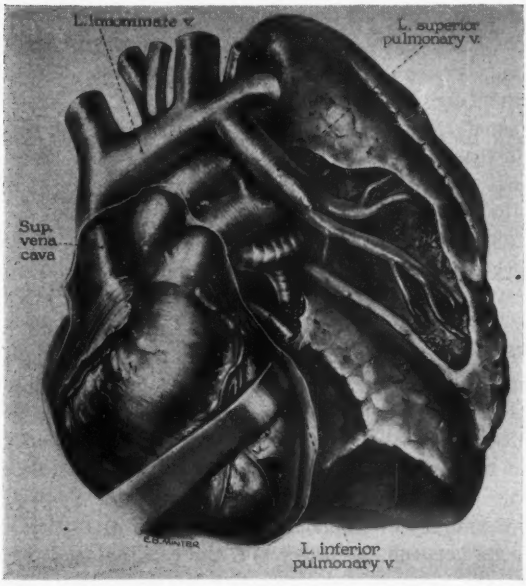

Fig. 4.—An anomalous pulmonary-systemic connection with the left superior pulmonary vein draining into the left innominate vein (white man, aged 40 years).

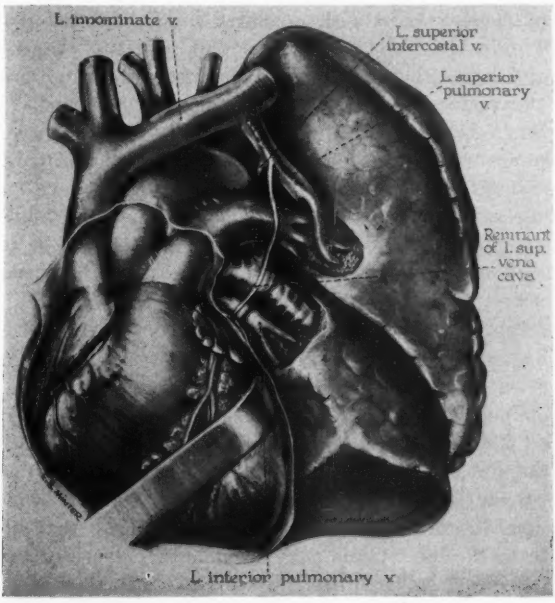

Fig. 5.—Drainage of the left upper lobe into the left innominate vein. Note the junction of the left superior intercostal vein and of the remnant of the left superior vena cava with the left superior pulmonary vein.

The embryology underlying the anomalous connections between the pulmonic and systemic venous systems has been discussed by many investigators. Brown,[6] in a study of the development of the pulmonary veins, described a plexus of capillaries surrounding the intestinal tract of the embryo which he termed the "splanchnic plexus" (Fig. 6,*A*). The cephalic portion of this capillary network drains into the anterior cardinal veins; the caudal portion drains into the posterior cardinal veins and the omphalomesenteric veins (Fig. 6,*B*). These latter veins are destined to become the unpaired portal vein.

With the development of a phylogenetically newer structure—the lung bud—a secondary plexus develops as an extension from the cephalic portion of the splanchnic plexus. This pulmonary plexus (Fig. 6,*C*) will have connections, therefore, with the splanchnic plexus, the cardinal veins, and the omphalomesenteric veins. The capillaries of the pulmonary plexus coalesce to form a longitudinal vein lying at the ventral pole of the pulmonary anlage. This vein passes through the dorsal mesocardium to open into the sinus venosus cephalad to and between the ducts of Cuvier. This common pulmonary vein will give rise to the four pulmonary veins found in the adult which enter the left atrium (Fig. 6,*D*). Most connections between the primary or splanchnic plexus and the secondary or pulmonary plexus are usually obliterated; as well as the connections between the latter and the cardinal and omphalomesenteric veins. Theoretically, any of these vascular channels may persist after birth.

Pertinent here are the observations of Zuckerkandl[23] made in 1881. Investigating the relationship of the pulmonary with the systemic venous system by injecting the pulmonary veins with a colored mass, he noted that the colored mass filled not only the pulmonary veins, but also the bronchial veins and through these the azygos, hemiazygos, and the mediastinal network of veins, all of which are derivatives of the cardinal veins. He claimed, therefore, that certain of the connections of the pulmonary plexus in the embryo, as described previously, may normally persist in the adult. The extent of these anastomotic channels in the adult varies with the individual; but regularly the pulmonary veins anastomose with the mediastinal plexus. The anomalous pulmonary-systemic connections reported in this paper may accordingly be regarded as an overgrowth of one of the normal connections as described by Zuckerkandl (Fig. 6,*E*), with a corresponding underdevelopment in the pulmonary plexus where the pulmonary vein in question should normally develop.

In many instances the pulmonary veins are normal in position at the hilus of the lung, yet enter the right side of the heart instead of the left. This may readily be explained on an embryologic basis. As previously mentioned, the original single pulmonary vein drains into the sinus venosus. With the displacement of the sinus to the right, the pulmonary orifice empties into the left portion of the primitive atrium just to the left of the left sinus valve. The formation of the interatrial septum definitely assigns the opening of the pulmonary vein to the left atrium. In an abnormal developmental procedure, the pulmonary orifice may be shifted to the right and thus account for many of the cases of aberrant drainage of the pulmonary veins into the coronary sinus, right atrium, superior vena cava, and inferior vena cava.

SIGNIFICANCE OF ANOMALOUS PULMONARY VEINS

Abnormalities in the number of pulmonary veins are of little clinical significance. Surgically, like abnormalities elsewhere in the body, one should be cognizant of these variations. When the veins are reduced in number, as in the case with a common pulmonary vein, the operative procedure may be simplified. The presence of multiple venous channels, however, may add further difficulties to an already tedious operative procedure.

Anomalous pulmonary-systemic connections do have important clinical significance at present, and may have even more in the future. When there is a complete drainage of the pulmonary circuit into the systemic system, the clinical picture, according to certain investigators, may be quite typical. The fact that an actual diagnosis of this condition during life has been made in only one

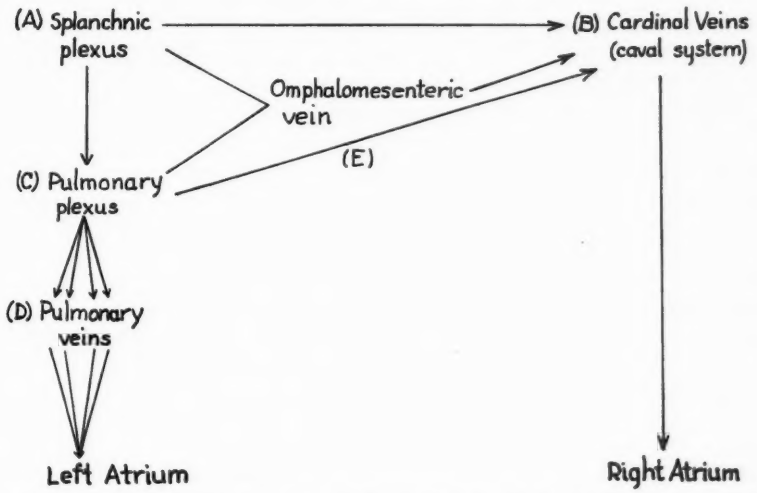

Fig. 6.—Diagrammatic representation of the connections of the pulmonary venous system in the embryo. (See text for explanation.)

instance[20] shows clearly that the anomaly has not been sufficiently emphasized in the past. In the near future, further cases will undoubtedly be detected with the aid of modern diagnostic methods of cardiac catheterization and angiocardiography. By these means, one may detect certain changes which are commonly encountered in this malformation, such as a high oxygen saturation of the blood in the right atrium compared with a decreased saturation of the peripheral blood, an increased right atrial pressure, an increase in the size of the right side of the heart, and a dilatation of the superior vena cava (the latter will occur, of course, only if the anomalous vessel drains into the superior vena cava or its tributaries). Electrocardiographic studies exhibit a right axis deviation. With all of these changes there is usually little or no cyanosis, except when accompanied by other congenital anomalies. Because of the pulmonary

congestion which frequently occurs due to increased pulmonary circulation, these cases are often interpreted as pulmonary disease rather than cardiac in origin. The recirculation also places extra strain upon the right side of the heart leading eventually to a right-sided heart failure.

As previously stated, the average age of the majority of these cases was 5.8 months. An operative correction of this deformity to prolong the life of children afflicted with it would certainly be a beneficial one, but thus far such a procedure has not been devised.

In cases where there is an incomplete drainage of the pulmonary system into the systemic system, the clinical signs are by no means typical. Indeed, a case may be asymptomatic depending upon the percentage of pulmonary tissue thus drained. Hughes and Rumore[14] claim that if less than 50 per cent of the pulmonary circuit drains into the right side of the heart, life to adulthood is possible. Those cases of incomplete drainage which are symptomatic may also be corrected surgically. There is only one report in the literature[7] which was reviewed by us, in which there was an attempted surgical correction of this malformation, and this was by ligation of the anomalous vessel. This attempt, however, was not successful. Brantigan[4] suggests either removal of that portion of the lung drained by the anomalous vessel, thereby preventing a right sided heart failure, or better, the malformation may be corrected by anastomosing the anomalous vessel to the left atrium either directly or indirectly.

Aside from the fact that incomplete drainage of the pulmonary into the systemic venous system may give rise to clinical signs and symptoms, it has importance in other ways. Many investigators have emphasized the high incidence of pulmonray disease in lungs which possess certain congenital anomalies. Conant and associates[8] described a case in which a patient had tuberculosis of the left lung. This lung was drained by a common vein which emptied into the left innominate vein. The functional value of this lung was reduced not only by the disease present, but, in addition, by the anomalous drainage of this lung into the systemic system which also increased the work load upon the right side of the heart. When spread of the disease process to the right lung occurred, death shortly ensued.

Such anomalous connections have surgical importance as well. For example, failure to recognize the drainage of a superior pulmonary vein into the systemic venous system might prove fatal when both lower lobes are removed for bronchiectasis.

Within the past two years, sixteen cases have been reported by nine different authors[2, 7, 9, 11, 12, 15, 17, 18, 21] in which anomalous pulmonary veins were diagnosed during life by means of cardiac catheterization, angiocardiography, and roentgenographic methods. All of these cases exhibited an incomplete drainage of the pulmonary circuit. It still remains for us, however, to diagnose those more important cases in infants having complete drainage of the pulmonary veins into the right side of the heart. Neverthless, these recently reported cases illustrate the fact that anomalous pulmonary veins can be diagnosed in living individuals, and offer assurance that they will be diagnosed more frequently in the future than they have been in the past.

SUMMARY

1. A general anatomic survey of anomalous pulmonary veins is presented; cases previously reported in the literature have been classified, and their incidence cited.

2. Three cases of anomalous-pulmonary-systemic connections observed by the author are described.

3. The clinical and surgical significance of anomalous pulmonary veins are briefly discussed.

ADDENDUM

Since submitting this paper, we have observed one case exhibiting a complete anomalous drainage of the pulmonary veins into the right side of the heart, and have collected four additional cases from the literature. In regard to the incomplete type of anomalous drainage, we have seen two cases and found two others reported within the past year. In addition, twelve cases have been recorded which were diagnosed during life by means of cardiac catheterization and/or angiocardiography.

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BRONCHOGRAPHY IN BRONCHIAL CANCER AND COLLATERAL VENTILATION

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GRONINGEN, HOLLAND

The following three case histories are of practical and theoretical importance.

CASE 1.—A man, aged 50 years, had been troubled with a bad cough five months previous to examination. His complaints completely disappeared, but three months previously he had felt a slight pain in the right side of his chest. He did not expectorate any blood. He felt tired, had little appetite, and lost 3 kilograms in weight in a few months' time. For this reason his physician sent him to the tuberculosis center. On examination, percussion and auscultation of the chest revealed no anomalies. A roentgenogram taken at this time showed an obvious anomaly, viz., an irregular shadow around the right hilus.

From the planigrams of the chest it appeared that the shadow around the hilus consisted of a conglomeration of globular shadows. On a section 16 cm. from the back, the impression was that the anterior side branch of the upper-lobe bronchus on the right, which lay within this mass of shadow, was completely occluded.

For bronchography the right side only was filled with 10 c.c. of lipiodol. The three usual photographs were taken, the dorsoventral one, the lateral one, and the oblique one. On all three photographs a stop was visible in the anterior side branch of the upper-lobe bronchus. Fig. 1 shows the dorsoventral photograph. The shadow around the hilus immediately strikes the eye. The right upper-lobe bronchus looks slightly compressed. The stop in the anterior side branch (*3*) is indicated by an arrow. Fig. 2 is the lateral photograph. On this one, too, the shadow is clearly visible. The anterior side branch is completely absent on this bronchogram, whereas all other side branches are well filled. Further toward the periphery in the anterior segment of the upper lobe not a sign of a shadow is visible.

When bronchoscopy was performed the right upper-lobe bronchus was immediately aimed at. The spur of the upper-lobe bronchus seemed slightly thickened. With the telescope the bifurcations were visible. The entrance of the anterior side branch was considerably swollen. The secretion was removed by suction for bacteriologic and cytologic examination (Keuning's opinion was: suspected carcinoma). Next the upper-lobe bronchus was focused at its maximum. With the curette, some tissue from the anterior side branch was removed. The microscopic diagnosis (Vos) was: carcinoma solidum. After complete functional examination in the internal clinic (Orie), pneumonectomy was performed by Eerland with favorable results to the present time. During the operation many carcinomatous glands were found near the hilus, which were also removed as far as visible and palpable.

A few points from this case history deserve closer consideration. A striking feature is the vague anamnesis. The complaints of pain in the chest were the most preponderant ones. This is a symptom that we have also often seen in cases of bronchial carcinoma. In this patient it was probably due to irritation of the pleura.

From a bronchologic point of view, two points are of special importance. This patient is a typical example of the success of what we nowadays call:

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Received for publication Sept. 4, 1951.

directed bronchoscopy. On the grounds of a localization, accurately determined by x-ray examination (planigraphy and bronchography) a certain bronchus is directly aimed at, and here some tissue is blindly removed by means of a curette. This directed bronchoscopy, as also indicated by Soulas and Versteegh from Bronkhorst's Utrecht school, is an important addition to the diagnostics. In a large number of cases of bronchial carcinoma the bronchoscopy is very simple. The tumor is immediately visible, and biopsy can easily be performed. Difficulties are chiefly met with when the localization is in the upper lobe and

Fig. 1.—Case 1. Dorsoventral bronchogram, right side.

more toward the periphery in the middle and lower lobes. In these cases one can reach further in this way. As a rule we perform this blind biopsy with a small curette. Its importance is sufficiently clear from the results obtained from a continuous series of 160 patients with bronchial carcinoma seen in the clinic during the last few years.

Total number of patients	160	
Bronchography	110	
Bronchoscopy	154	
Microscopy +	123	= 80 per cent
Biopsy +	106	
Curettage +	17	

As appears from these statistics, bronchoscopy was performed on nearly all patients. In six cases the diagnosis could also be established with certainty without it, e.g., by means of an excision from supraclavicular glands. On most patients bronchography was performed. This is chiefly done for two reasons. First with an eye to the "directed bronchoscopy." For this reason bronchography is usually performed before bronchoscopy. Second, the bronchogram often shows various anomalies, that are sometimes of great importance, e.g., in determining the operability. The second case clearly demonstrates this fact.

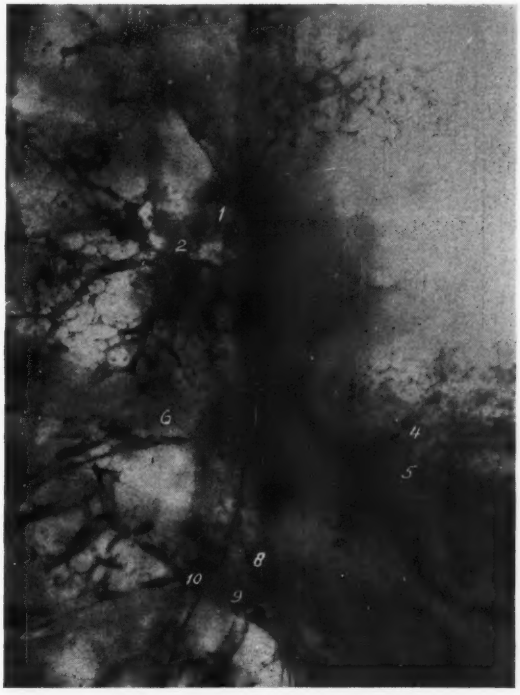

Fig. 2.—Case 1. Lateral bronchogram.

The result that in 80 per cent of these cases it was possible to establish a certain diagnosis by means of bronchoscopy is very satisfactory. This high percentage was also found of late years at Utrecht by Versteegh and Swierenga and, among others, by Holinger and Soulas. This is only due to the improved technique of the last few years. Until recently the usual percentages given were 50 to 60 per cent. It appears that an important addition is furnished by the blind biopsy, 17 times on 123 patients. It seems fairly certain that this percentage will not increase in the future, on the contrary, it will decrease. Lately we see more and more difficult cases with tumors in the periphery. This is chiefly a result of the ever-increasing group fluoroscopy of the population of

Holland. Fortunately, we now have cytology at our disposal, which was introduced into bronchology by Clerf and Herbut. The Groningen results are in the main very good, thanks to the cooperation with the histologist Keuning. In these cases it is also important to perform directed bronchoscopy in order to remove the secretion from the suspect bronchus.

In a quite different respect this case is also important. The shadow, visible in the right half of the chest, is completely caused by the tumor and the metastases in the hilar glands. There is no shadow as a result of anomalies in the pul-

Fig. 3.—Case 2. Dorsoventral bronchogram, right side.

monary tissue of the anterior segment of the upper lobe. This fact is remarkable. A big side branch completely, or nearly so, occluded has still not resulted in atelectasis. In these cases the possibility should always be taken into account that ventilation is still possible alongside the obstruction. This fact was called attention to by my friend Bronkhorst by a demonstration of two evident cases. Personally, I have always seen atelectasis in cases of a complete stop of a main or lobar bronchus. Atelectasis was often seen even when lipiodol did still pass through the stenosis. So this is just the reverse. It is more plausible to explain the fact that an occluded part of the lung contains air, by ventilation by way

of the pulmonary tissue from out of the other segments of the upper lobe. This is collateral ventilation, to which particular attention has been paid in Groningen, thanks to the cooperation of Dirken.[1] It was first described by van Allen and his collaborators.[2]

CASE 2.—A man, aged 59 years, had had his complaints for a short time only that he had had a bad cough. The internist found no anomalies when examining the chest, but on the roentgenograms he found a broadened hilus, particularly on the right. Fig. 3 also clearly shows this shadow. But there is no shadow in the lower part of the lower lobe, the space

Fig. 4.—Case 2. Lateral bronchogram.

between heart and diaphragm is occupied by air-containing tissue. Fig. 3 shows the dorso-ventral bronchogram. This reveals a number of anomalies. In the first place the carina is considerably broadened (*a*). The right upper-lobe bronchus is well filled, the three segmental bronchi show no anomalies. Somewhat lower down the stem bronchus suddenly grows much narrower with a quite smooth margin, apparently the bronchus is compressed from the medial side (*b*). On the place of the bifurcation of the middle- and lower-lobe bronchi medially a small irregularity is visible. One has some difficulty in further finding one's way in the bunch of downgoing bronchi. The lateral (*4*) and the medial (*5*) branches of the middle lobe bronchus are still recognizable with fairly great probability. Behind these, however, there are two more descending bronchi that cannot be explained as basal branches. The lateral photograph gives the solution. Above, one sees the main bronchus with the entrance of the upper-lobe bronchus. The basal bronchi are completely absent, in the lower-lobe

bronchus there is an obstruction with an irregular margin. Only the middle-lobe bronchi (*4* and *5*) and the apical branch of the lower lobe (*6*) are filled. The entrances of both bronchi, but particularly that of the middle lobe, are slightly narrowed. The two descending branches on the dorsoventral bronchogram of Fig. 3 are, therefore, side branches of the apical bronchus of the lower lobe. The occluded part of the lung contains the four basal

Fig. 5.—Case 3. Dorsoventral bronchogram, right side.

segments. This part of the lung is smaller than normal, for the middle lobe covers the anterior part of the diaphragm. Apparently there is hypoventilation in the occluded part of the lung, but, thanks to the collateral ventilation, it still contains air. Owing to this the physical and x-ray examinations of the chest revealed no phenomena of occlusion of the lower-lobe bronchus.

Bronchoscopy revealed no anomalies of the trachea. The carina was considerably broadened. The stem bronchus was narrowed. Immediately below the bifurcation of the middle-lobe bronchus and the apical branch of the right lower lobe the bronchus was completely occluded by easily bleeding granulations. Biopsy revealed a planocellular carcinoma. Owing to the bronchologic examination it was, therefore, possible not only to establish the diagnosis with certainty, but it was also apparent that the patient could not be operated upon owing to the extension into the mediastinal glands (broadened carina and pressure on the stem bronchus).

Fig 6.—Case 3. Lateral bronchogram.

CASE 3.—A 52-year-old man had a productive cough for a few months, with no hemoptyses. On the right side of the heart a perihilar shadow was visible. This shadow, which is very suspect for atelectasis of the middle lobe, is clearly visible on the bronchogram of Fig. 5. Fig. 5 is a dorsoventral photograph, showing that the right lower lobe contains a fair quantity of air. The space between heart and diaphragm is not portrayed. In this photograph the obstruction is clearly visible; it has a conical shape. The impression is conveyed that an occlusion exists in the stem bronchus (with this term is meant, in Groningen, the main bronchus of the right side below the bifurcation of the upper-lobe bronchus, so the common bronchus of the middle and lower lobes). Fig. 6 is the lateral photograph, demonstrating once more, just as in the preceding case, the importance of taking photographs in various directions, also in bronchography. The picture thus obtained is quite different. There is not one obstruction, but two: an occlusion of the middle-lobe bronchus and an occlusion of the lower lobe bronchus below the apical branch (*6*). With bronchoscopy a tumor, occluding the lower-lobe bronchus below the branching off of the apical branch, was found. Biopsy revealed a carcinoma.

This case is particularly demonstrative, as it so clearly shows the difference between the occlusion of the middle-lobe bronchus, in which case collateral ventilation is impossible, resulting in atelectasis, and the occlusion of the lower-lobe bronchus, in which case collateral ventilation is still possible through the apical branch (*6*), thus probably prohibiting atelectasis. From the surgical preparation it was stated that the occlusion in the lower lobe was caused by the primary tumor and the occlusion in the middle lobe by glandular metastases.

Naturally these cases are exceptions in the great number of patients suffering from bronchial carcinoma now seen in the clinic. They are, however, of great theoretical importance. An important point is that very considerable bronchial stenosis may exist, whereas an examination, also with roentgenograms, does not reveal any signs of bronchial stenosis. This is in all probability caused by collateral ventilation. This condition can only have a temporary existence. For, as was already described by van Allen, an inflammation easily eliminates the collateral ventilation. Bronchial stenosis is nearly always accompanied by inflammation. Further growth of the tumor, moreover, will after some time obturate the apical branch from the lower lobe thus certainly eliminating collateral ventilation.

In the preceding communication,[1] the importance of collateral ventilation in cases of foreign body was pointed out. We saw several examples of it. The above cases make it clear that collateral ventilation is also important in cases of tumors.

SUMMARY

Three cases of bronchial carcinoma are described, in which probably collateral ventilation existed. In cases of bronchial carcinoma bronchoscopy is as a rule preceded by bronchography. Bronchography can often furnish important indication, also in cases of carcinoma, e.g., concerning the operability. It is of particular importance with tumor in the periphery for the performance of a directed bronchoscopy. Its importance is demonstrated by the statistics of our last 160 cases in which, thanks to this directed bronchoscopy, a positive biopsy was possible in 80 per cent.

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PHYSIOLOGIC STUDIES FOLLOWING THORACIC SURGERY

I. Immediate Effects of Thoracoplasty

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INTRODUCTION

THIS is the first of a series of reports dealing with the physiologic effects of thoracic surgical procedures employed in the treatment of cavitary pulmonary tuberculosis. The effects produced by lobectomy, pneumonectomy, and segmental resections will be covered in subsequent communications. In the choice of a surgical procedure suitable for a given patient three factors must be considered: (1) The nature of the tuberculous lesion and the merits of the various surgical approaches as definitive treatment for the lesion at hand[1]; (2) the pulmonary reserve of the patient and his ability to withstand the permanent loss of function following the operation; and (3) the magnitude of the immediate stress which attends the procedure, and the ability of the patient to withstand this stress. With these considerations, the procedure is then designed to fit both the lesion itself and the patient as a whole. It was felt that a careful study of the physiologic effects of the several surgical approaches to the treatment of cavitary pulmonary tuberculosis would, therefore, be of value.

Excisional surgery is rapidly gaining in popularity in the treatment of tuberculosis. Much is being written upon the specific indications for resection and upon its efficacy in the treatment of the various types of tuberculous lesions. In the presence of normal pulmonary reserve, most surgical procedures present little difficulty, but in many cases diffuse pulmonary disease (emphysema and fibrosis) coexists with the tuberculous lesion under attack. It is this coexisting disease that frequently impairs the over-all pulmonary function and is, therefore, the determining factor in the patient's ability to withstand major thoracic surgery.[1, 2] A great deal of experience is needed in assessing the ability of a borderline case to withstand the resection of pulmonary tissue. Thus, in selecting the ideal operation for the treatment of a given tuberculous lesion, a compromise must frequently be made in order to keep the immediate surgical trauma and ultimate loss of pulmonary function within the limits imposed by the patient's over-all pulmonary reserve. The purpose of this series of reports has been to quantitate and compare the immediate stress and the ultimate sacrifice of pulmonary function produced by the various operations currently in use in the treatment of cavitary pulmonary tuberculosis.

The present report is concerned with the physiologic effects that were noted within twenty-four hours of each stage of the three-stage thoracoplasty.

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Published with approval of Chief Medical Director. The statements and conclusions published by the authors are the result of their own study and do not necessarily reflect the opinion or policy of the Veterans Administration.

Received for publication Oct. 15, 1951.

Thoracoplasty was chosen for the first study because it is a well standardized and familiar procedure in thoracic surgery. The magnitude of the stress imposed by thoracoplasty is so well known to chest surgeons that it may serve as a yardstick for the quantitative comparison of the effects of thoracic operations of greater magnitude.

The observations herein reported may be divided into two parts: (1) The preoperative determination of pulmonary function, and (2) the evaluation of the physiologic status of the patient sixteen to twenty hours postoperatively. The preoperative study served two purposes: (a) to aid in the objective estimation of the pulmonary reserve, and (b) to furnish a base line for immediate postoperative observations and for subsequent analysis of the ultimate functional changes to be reported at a later date.

METHODS

A few days prior to the first stage of thoracoplasty, the patient was taken to the laboratory at 7:30 A.M. in the fasting state. Weight, height, age, and vital capacity were recorded. The functional residual volume and index of pulmonary mixing were determined in duplicate in the manner described by Cournand and associates.[3] The subdivisions of the vital capacity were measured from the tracing made on a standard Collins or McKesson metabolimeter. This first group of tests served to acquaint the subject with the respiratory apparatus, in order that he might be more relaxed during subsequent observations.

The following morning the patient was brought to the laboratory in the fasting state. The oral temperature was recorded. A large bore needle (Cournand modification of the Linde needle) was inserted into the femoral artery under local anesthesia. This needle was left in place throughout the entire procedure.

The expired air was then collected for a seven-minute period in the Tissot gasometer, which had previously been thoroughly rinsed with the patient's expired air. An open circuit was used, the patient inhaling room air through a valve of 53 c.c. dead space. During this period the pulse and respiratory rates were recorded, and two samples of arterial blood were withdrawn through the Cournand needle without disturbing the patient. Resting ventilation and basal metabolic rate were determined by measurement and quantitative analysis of the expired gas collected in the Tissot gasometer. The physiologic dead space and alveolar ventilation were also calculated from these data (see below).

Next, the subject inhaled 100 per cent oxygen through a similar open circuit. After a period of at least seven minutes, a third sample of arterial blood was withdrawn, and the needle was removed. This sample was analyzed at once for oxygen content to determine the amount of oxygen physically dissolved in the blood. This value was used in the calculation of the pulmonary shunt, after the method of Fasciolo and Chiodi,[4] using the formula:

$$X = \frac{V a_{O_2} - V v_{O_2}}{V P a_{O_2} - V v_{O_2}} \times 100$$ where: X is per cent of blood passing through aerated lung tissue;

$V a_{O_2}$ is arterial blood O_2 content of patient; $V v_{O_2}$ is venous blood O_2 content (actually in this study $V a_{O_2} - V v_{O_2}$ was not determined, but a value of 4 volumes per cent was assumed instead); $V P a_{O_2}$ is arterial hemoglobin capacity plus the mean value for dissolved O_2 found in a series of normal subjects. This is taken as the theoretical normal for this patient if he had only a normal amount of pulmonary shunt.

With the patient in a sitting position, the maximum breathing capacity was measured directly from the volume of air collected in a Douglas bag during thirty seconds of maximal voluntary hyperventilation. The resistance in the valve system was minimal at the rate of ventilation achieved by most of our subjects.

The blood for the determination of the pH was drawn into a 10 c.c. syringe which had been warmed to 37° C. It was agitated briefly with a small amount of mercury and 0.1 c.c. heparin, and the pH determination carried out within five minutes at 37° C. in the manner described by Wilson and Ognanovich.[5] In a series of normal subjects the mean pH by this method was 7.40 ± 0.02.

The arterial blood carbon dioxide content was determined immediately in duplicate after the method of Van Slyke and Sendroy.[6] The oxygen content and hemoglobin capacity were determined in duplicate by the method of Van Slyke and Neill.[7] Originally a correction was made for change in hemoglobin concentration which occurs in the tonometer in the manner described by Comroe and Walker[8] and a 2 per cent higher saturation found by this method. This was felt to contribute nothing, and so values are expressed in the conventional manner. The normal value in this laboratory is 96.50 per cent uncorrected and 98.50 per cent corrected for the change in hemoglobin concentration.

The oxygen content and capacity of the blood collected during the breathing of pure oxygen were analyzed. The oxygen content of the blood collected during the breathing of 100 per cent oxygen, minus the hemoglobin capacity, was taken as an estimate of the oxygen which was physically dissolved in the blood. The normal value for the dissolved oxygen under these conditions is 1.80 ± 0.294 volumes per cent in this laboratory. The true value should be 2.04 volumes per cent under these circumstances if all factors are taken into account.[9] This method, however, does give a value which is fairly constant in normals and in the patients preoperatively. The fact that little blood passes through the diseased portion of the lung has been observed by Berggren[10] and also by Björk.[11]

In determining the maximum breathing capacity, the thirty-second period was chosen because it would give sufficient gas for accurate measurement and would at the same time avoid gross fatigue on the part of the patient. The normal values in this laboratory are somewhat lower than those given by Cournand and Richards.[12] This is probably because the method used by these investigators involved the rebreathing of expired air in a spirometer, and the method employed here utilized fresh room air for each breath. In this manner, the additional stimulus of increased carbon dioxide and decreased oxygen content of the inspired gas was avoided. Parallel determinations in this laboratory using both methods showed that the spirometric method of Cournand gave consistently higher values.

Arterial blood P_{CO_2} (Pa_{CO_2}) was estimated from the values for arterial blood CO_2 content and pH. The normogram prepared by Singer and Hastings[13] was used for convenience. Alveolar P_{CO_2} ($P_{A_{CO_2}}$) was assumed to be equal to arterial blood P_{CO_2} after the suggestion of Riley.[14] CO_2 excretion per minute ($\dot{V}_{CO_2}$) was determined directly by measurement and analysis of the expired air. Alveolar ventilation per minute was calculated by use of the formula:[14]

$$\dot{V}_A = \frac{\dot{V}_{CO_2}}{F_{A_{CO_2}}}$$

where: $\dot{V}_A$ = alveolar ventilation in cubic centimeters per minute.

$\dot{V}_{CO_2}$ is rate of CO_2 excretion in cubic centimeter per minute.

$F_{A_{CO_2}}$ is fraction of alveolar gas made up by carbon dioxide.

$\dot{V}$ is resting minute volume of ventilation in cubic centimeters per minute at standard condition of temperature and pressure.

Alveolar ventilation per breath was calculated by use of the formula:

$$V_A = \frac{\dot{V}_A}{f}$$

where: V_A is alveolar ventilation per breath.

f is respiratory rate per minute.

TABLE I

	MEAN NORMAL	MEAN PREOPERATIVE	MEAN POSTOPERATIVE NO. 1	MEAN POSTOPERATIVE NO. 2	MEAN POSTOPERATIVE NO. 3
Oral Temp. (°F.)		98.3 ± .4	100.1 ± .8*	99.2 ± .7*	99.6 ± .7*
Pulse Rate/Min.		81. ± 10.2	108. ± 13.*	108. ± 10.*	113. ± 12.*
Metab. Rate (Cal/hr.)/M^2	35.0 ± 2.7	39.2 ± 3.2	45.7 ± 2.8*	44.1 ± 2.6*	49.3 ± 8.4*
Resp. Quotient	.90 ± .04	.87 ± .08	.76 ± .03*	.80 ± .06*	.78 ± .08*
Arterial Blood:					
pH	7.40 ± .01	7.40 ± .01	7.40 ± .01	7.40 ± .01	7.39 ± .01
CO_2 cont. MM/L	21.3 ± 1.0	21.3 ± 1.1	21.4 ± .5	21.3 ± .9	21.5 ± 1.6
P_{CO_2} (mm. Hg)	39.0 ± 2.0	40.6 ± 2.5	41.7 ± 3.0	40.6 ± 2.0	41.4 ± 4.1
Hgb sat'n (%)	96.5 ± .8	95.7 ± 1.7	90.3 ± 2.1*	91.1 ± 1.5*	90.7 ± 2.9*
Pulm. Shunt (%)	0	0.5%	14.4%*	17.0%*	14.5%*
$\dot{V}$ (c.c./min.)	5641. ± 601.	6180. ± 892.	7058. ± 856.*	7543. ± 1190.*	7147. ± 1302.*
f (Resp./min.)†	12. ± 1.8	14.5 ± 1.7	18.9 ± 3.8*	18.8 ± 2.6*	21.0 ± 9.4*
V_T (c.c.)	499. ± 58.	430. ± 83.	379. ± 66.	407. ± 58.	384. ± 103.
V_D (c.c.)	144. ± 32.	127. ± 49.	125. ± 36.	144. ± 47.	135. ± 33.
$\dot{V}_D$ (c.c./min.)	1605. ± 421.	1828. ± 700.	2355. ± 733.*	2719. ± 919.*	2786. ± 619.*
V_A (c.c.)	310. ± 47.	257. ± 59.	207. ± 36.*	211. ± 27.*	203. ± 63.*
$\dot{V}_{CO_2}$ (c.c./min.)	212. ± 23.	217. ± 37.	226. ± 24.	227. ± 33.	237. ± 67.
$\dot{V}_A$ (c.c./min.)	3496. ± 381.	3685. ± 764.	3835. ± 500.	3961. ± 675.	4204. ± 1448.
Vital Capacity c.c./M^2	2290. ± 274.	1820. ± 360.†			
Resid. Volume c.c./M^2	680. ± 148.	1070. ± 480.†			
Reserve Volume c.c./M^2	462. ± 116.	355. ± 150.			
Total Lung Vol. c.c./M^2	2970. ± 313.	2950. ± 570.			
$RV/_{TLV}$ %	23.0 ± 4.1	35.9 ± 12.4†			
Index Pulm. Mixing (% N_2)	1.45 ± .23	2.83 ± .88†			
Max. Br. Cap. L/min./M^2	61. ± 6.	41. ± 12.†			

*Statistical analysis reveals this mean to differ significantly from the preoperative mean ($p = 0.05$ or less).

†Statistical analysis reveals this mean to differ significantly from the mean value obtained in sixteen normal subjects ($p = 0.05$ or less).

Physiologic dead space was calculated by use of the formula:

$$V_D = V_T - (V_A + DS_I)$$

where: V_D is physiologic dead space in cubic centimeters.
V_T is tidal volume
V_A is alveolar ventilation per breath
DS_I is dead space of respiratory valve (53 c.c. which when corrected to BTPS is 46 c.c.)

The dead space ventilated per minute was calculated by use of the formula:

$$\dot{V}_D = V_D \times f$$

where: $\dot{V}_D$ is cumulative volume of dead space ventilated in cubic centimeters per minute.

These calculations, although apparently complex, yielded the most useful information obtained from this study for understanding the physiologic changes which follow surgical procedures involving the thoracic cage.

The postoperative observations were made in the patient's room at 7:30 A.M., on the morning following surgery. These consisted in the recording of the temperature, pulse, respiratory rate, and vital capacity. The expired air was collected and arterial blood was drawn in the same manner as described above. The oxygen dissolved in the blood during the inhalation of 100 per cent oxygen was again determined. Each patient was studied in the fasting state.

CLINICAL MATERIAL

Ten male patients, of ages 23 to 62 years, were studied. Each had previously been reviewed and accepted for thoracoplasty by members of the medical and thoracic surgical services. Each patient had predominately unilateral, upper-lobe, cavitary pulmonary tuberculosis. The contralateral lung was either clear or contained apparently stable tuberculous lesions. There was no significant pleural involvement. Clinical observations showed the patients to vary from men who were physiologically normal to men who were dyspneic on slight exertion.

Normal values for this laboratory were determined by testing sixteen normal young males. These figures were used as a standard by which to judge the pulmonary function of the patients before surgery.

Table I gives the mean values for the preoperative tests in this group of patients, and the mean value found in the group of normal subjects.

It can be seen that the index of pulmonary mixing, and residual volume were significantly greater in this group of patients than in the normal subjects. Several of the patients actually had moderately severe pulmonary emphysema with a residual volume greater than 40 per cent of the total lung volume. The mean vital capacity and maximum breathing capacity were significantly lower than in the normals. The resting minute volume of ventilation, basal metabolic rate, and ventilatory equivalent for oxygen did not differ significantly from the normal values. The arterial blood oxygen saturation, carbon dioxide content, carbon dioxide tension, and pH were also within normal limits. The mean for the oxygen dissolved in arterial blood by breathing 100 per cent oxygen was not significantly different from the normal. This indicated that there was no significant pulmonary shunt in these patients.

*Statistical analysis reveals this mean to differ significantly from the preoperative mean ($p = 0.05$ or less).
†Statistical analysis reveals this mean to differ significantly from the mean value obtained in sixteen normal subjects ($p = 0.05$ or less).

The technique of thoracoplasty used in the patients comprising this study was as follows: The first stage consisted of the removal of the first rib and posterolateral portions of ribs 2, 3, and 4. The second stage was done from twelve to sixteen days later and consisted of the removal of the anterior segments of ribs 2 and 3 and cartilages of 1, 2 and 3. At the third stage, performed twelve to sixteen days following the second, the posterolateral segments of ribs 5, 6, and 7 were removed. It is felt by the Thoracic Surgery Service that these three stages divide the surgical trauma into approximately equal parts. Local infiltration with 1 per cent procaine was the only anesthetic employed.

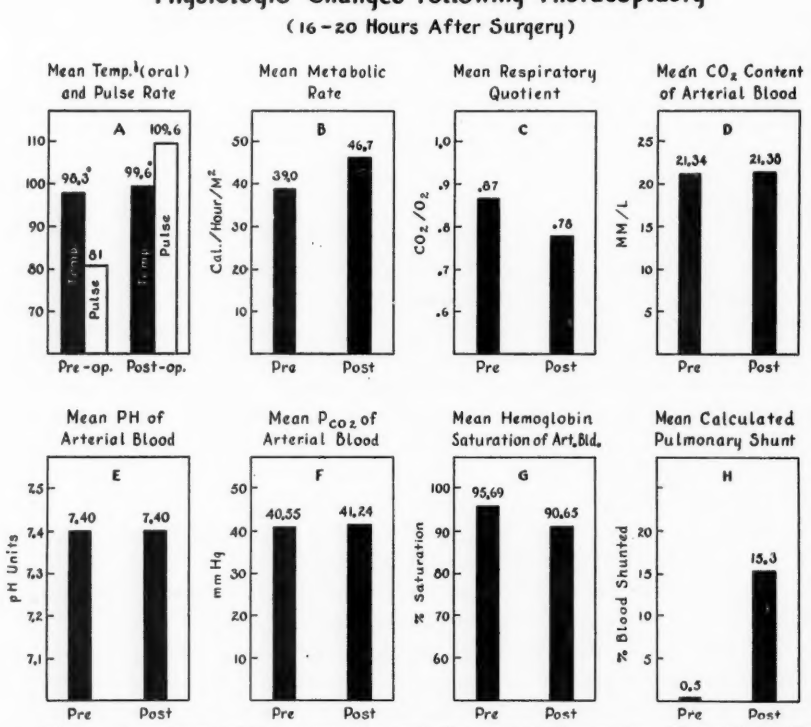

Fig. 1.—Comparison of the preoperative with the postoperative states. The postoperative figures represent the weighted means of the three postoperative determinations in each subject making a total of twenty-nine postoperative studies in the ten subjects. In one subject (M. J.) a pneumonectomy was performed at the time of the third stage thoracoplasty, and the third study is therefore not included in this report.

RESULTS

The mean physiologic effects of the individual stages are given in Table I. Detailed analysis of the results by stages revealed that there was no difference between the effects produced by the various stages of thoracoplasty. The results of the mean values by stages have, therefore, been averaged and compared with the preoperative control value. These comparisons are given in Figs. 1 and 2.

The mean oral morning temperature rose from 98.3°·F. preoperatively to 99.8° F. the day following surgery (Fig. 1*A*). This change, though slight, was statistically significant. The mean preoperative pulse rate was 81 per minute as compared with a mean of 109.6 postoperatively (Fig. 1*A*).

The mean carbon dioxide content of arterial blood in the preoperative period was 21.34 milliequivalents per liter and postoperatively was 21.38 milliequivalents per liter (Fig. 1*D*). The pH of the arterial blood remained un-

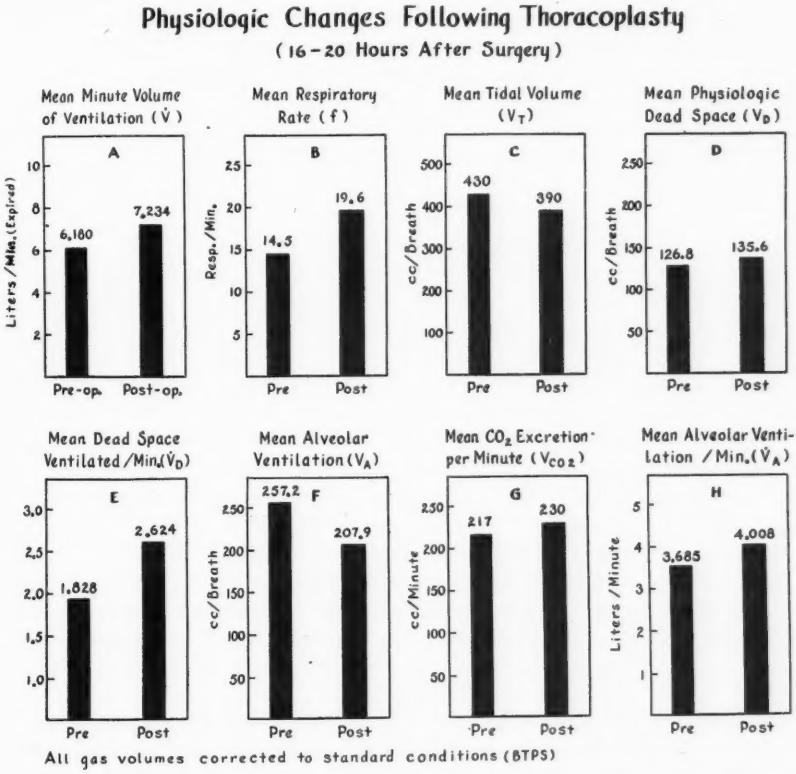

Fig. 2.—Comparison of the ventilatory mechanism of the preoperative and postoperative states. The postoperative figures represent the weighted means of the three postoperative determinations in each subject, making a total of twenty-nine postoperative studies in the ten subjects.

changed at 7.40 postoperatively (Fig. 1*E*) and the carbon dioxide tension rose only from 40.55 mm. Hg preoperatively to 41.24 mm. Hg the morning after surgery (Fig. 1*F*). None of these changes was of significance.

The mean arterial hemoglobin saturation in the first twenty-four hours after the surgical procedure fell to 90.31 per cent from the control mean of 95.07 per cent, while the patient was breathing ambient air (Fig. 1*G*). This change was of significance.

The quantity of oxygen physically dissolved in the arterial blood by the exposure to an alveolar oxygen tension of about 600 mm. Hg fell from 1.78 volumes per cent preoperatively to 1.09 volumes per cent postoperatively. From this one can calculate that 0.69 volumes per cent fall occurred in the oxygen content of blood leaving the lungs. This change represents a 15.3 per cent absolute shunt through nonventilated lung tissue (Fig. 1*H*).

The minute volume of ventilation rose from a preoperative mean of 6,180 c.c. per minute to 7,234 c.c. per minute the morning following surgery (Fig. 2*A*). This change was accounted for primarily by an increased rate of respiration from 14.5 to 19.6 per minute (Fig. 2*B*). The tidal volume fell from a mean of 430 c.c. preoperatively to a mean of 390 c.c. postoperatively (Fig. 2*C*). The volume of the physiologic dead space changed only from a mean of 126.8 c.c. preoperatively to 135.6 c.c. in the immediate postoperative period (Fig. 2*D*). The volume of dead space that was ventilated each minute (physiologic dead space times respiratory rate) rose from a mean of 1,828 c.c. per minute preoperatively to 2,624 c.c. per minute postoperatively, i.e., a mean individual increase of 58 per cent above the preoperative level (Fig. 2*E*). Alveolar ventilation per breath decreased significantly from 257.2 c.c. before surgery to 207.9 c.c. shortly thereafter (Fig. 2*F*). The total volume of carbon dioxide excreted by the lungs per minute rose only from 217 c.c. per minute preoperatively to 230 c.c. per minute after surgery (Fig. 2*G*), in spite of the fact that the mean metabolic rate rose 19 per cent following surgery (Fig. 1*B*). This apparent discrepancy is accounted for by a significant fall in the mean respiratory quotient from 0.87 preoperatively to 0.78 postoperatively (Fig. 1*C*). The lower respiratory quotient following surgery was probably due to the poor nutrition on the day of surgery, a period of about twenty-four hours immediately preceding the observation. The volume of alveolar ventilation per minute (alveolar ventilation times respiratory rate) was increased proportionately from a mean of 3,685 c.c. per minute before operation to 4,008 c.c. per minute following surgery (Fig. 2*H*). The change in carbon dioxide excretion and alveolar ventilation per minute did not prove to be of statistical significance.

DISCUSSION

The interest of this laboratory in the physiologic changes following thoracic surgery was aroused by the occurrence of unexplained shock in two patients early in 1949. The marked uncompensated respiratory acidosis which was found (arterial blood pH 7.10) was thought to be the basis for the clinical picture.

It is well known that many patients who require surgical therapy for pulmonary tuberculosis also have concomitant diffuse pulmonary disease such as emphysema and fibrosis.[2] The presence of an extensive diffuse pulmonary process of this type may lead to dyspnea during mild exertion or even at rest. Such a situation may preclude certain major operations on the chest. It was felt that an accurate determination of the stress placed upon the body in the postoperative period and of the efficiency with which the recently operated patient could meet this stress would be of value.

The foremost function of the lungs is the absorption of oxygen and the elimination of carbon dioxide. Of these, the carbon dioxide excretory factor is of the greater importance since oxygen is easily supplied therapeutically. The lungs are solely responsible for the elimination of carbon dioxide and thus play a vital role in the maintenance of the acid-base balance of the blood. There is as yet no way whereby the excretion of carbon dioxide can be facilitated save by an increased alveolar ventilation. Furthermore, for practical purposes, sole reliance for such ventilation must be placed upon the natural mechanisms of the body. If these mechanisms prove insufficient, carbon dioxide retention and respiratory acidosis ensue.

Two recent papers[16, 17] have called attention to the development of respiratory acidosis *during* intrathoracic operations. The authors felt that the carbon dioxide retention occurred because of insufficient alveolar ventilation during the procedure. This was largely a complication of the artificial respiration that is part of endotracheal positive pressure anesthesia. Since the present study was an attempt to define the physiologic effects of the surgical trauma itself and not of the anesthesia, a minimum of sixteen hours was allowed to pass after the completion of surgery before observations were made. This time interval was observed even in the patients undergoing thoracoplasty (in whom local anesthesia alone was employed) in order that the data would be comparable to the data collected upon patients in whom general anesthesia was to be employed.

No instance of carbon dioxide retention occurred in this series of cases studied after the three stages of thoracoplasty. The arterial blood pH and P_{CO_2} were unchanged from the control values. The normal P_{CO_2} in the postoperative period also indicates that the mild anoxemia which existed was insufficient to stimulate an increase in respiratory volume.

The degree of anoxemia which existed on the morning following surgery was slight, except in one case (G. J.) in whom atelectasis of the lower lobe occurred because of plugging of the bronchus by retained secretions. The fall from a mean of 95.79 per cent hemoglobin saturation to 90.66 per cent can largely be accounted for by the shunt of blood through recently collapsed lung tissue which was not being aerated. There was a 15.3 per cent shunt estimated to occur in the lung on the morning following surgery. The method used for this determination involved the breathing of 100 per cent oxygen by the subject and reflects only that blood passing through lung tissue which was not aerated at all. It does not demonstrate the quantity of blood passing through portions of lung which are ventilated to a less than normal degree. The effective shunt when the patient is breathing room air would, therefore, be even greater than these data demonstrate. The observation of a significant shunt of blood through recently collapsed pulmonary tissue is in agreement with the work of Berggren[10] in man and of Björk[11] in animals.

The observations herein reported throw light upon the respiratory adjustments made by the body after surgery to the chest. It has been shown in this series of cases that: (1) the total volume of ventilation per minute increased

appreciably; (2) the respiratory rate rose significantly; (3) the amount of carbon dioxide formed per minute rose slightly; (4) the respiratory quotient fell postoperatively.

From these facts certain deductions can be made relative to the physiologic mechanisms involved in the postoperative adjustment. The maintenance of alveolar ventilation was a necessary response to the continued formation of carbon dioxide at about the same or slightly higher level. If this had not occurred carbon dioxide retention and respiratory acidosis would have resulted. Pain limited the chest expansion possible per breath and thus decreased the tidal volume. A fall in tidal volume necessarily results in a decrease in alveolar ventilation achieved by each breath, since the volume of physiologic dead space cannot diminish. Actually, the physiologic dead space may be increased by rapid breathing because of the quicker movement of air through the respiratory bronchioles. In order to maintain adequate alveolar ventilation in the face of a smaller alveolar ventilation achieved by each breath, an increase in the rate of respiration occurred. Such an increase in rate with a smaller tidal volume constitutes an inefficient means of achieving adequate alveolar ventilation, since with every breath energy is wasted to move about 150 c.c. of air back and forth in the physiologic dead space. That this is of importance in the patients studied is shown by the great increase (58 per cent) in the volume of dead space ventilated per minute.

The data afford an explanation for Cournand's observation[2] that patients whose maximum breathing capacity was less than six times greater than their resting volume of ventilation (breathing reserve of less than 85 per cent) did not tolerate thoracoplasty satisfactorily. It has been shown that there is a definite and prolonged (several days) need for an increased minute volume of ventilation following thoracic surgery and that this must be accomplished by more rapid respiration. Patients who are unable to maintain this increased rate and volume of respiration would develop carbon dioxide retention which is a serious complication. A patient who is not able to achieve a sixfold increase over his volume of resting ventilation by a twenty- to thirty-second effort might well not be able to meet the prolonged need for a more rapid respiratory exchange after thoracic surgery.

It should be emphasized that the observations herein reported were made at a time when all direct effects of limited respiratory exchange during surgery would have been compensated and at a time when there was little increase in the rate of carbon dioxide excretion. The respiratory changes noted would, of course, be greatly exaggerated by the existence of acidosis with its attendant need for a greater alveolar ventilation. Such would be the situation in the respiratory acidosis at the close of intra-thoracic procedures, as observed by Beecher[16] and by Taylor.[17] The physiologic adjustments to such circumstances are presently being investigated and will be reported in a later paper.

From the studies reported in this paper it can be stated that if the P_{CO_2} is found to be high on the day following thoracoplasty a diagnosis of pulmonary insufficiency is justified and steps should be taken to increase the

volume of alveolar ventilation. This can probably best be done by the performance of a tracheotomy. By lowering the physiologic dead space this procedure allows a greater alveolar ventilation to be achieved by the same or smaller tidal volume of ventilation. The tracheotomy opening also presents less resistance to the exchange of air in and out of the lungs and, therefore, lessens the paradoxical motion of the flaccid chest wall.[18] Both of these effects help to produce a more efficient respiratory mechanism. The effect of tracheotomy in such cases is currently under investigation in this laboratory.

SUMMARY AND CONCLUSIONS

1. The immediate physiologic postoperative effects of three stage, seven-rib thoracoplasty were studied by stages in ten men ranging in age from 23 to 62 years. Careful preoperative evaluation of pulmonary function was done in each case. Clinically the patients varied from men who were normal to men who were dyspneic on slight exertion.

2. A significant, but rather mild, degree of anoxemia was found to follow surgery in each instance. In one case the anoxemia was more severe but in this one there was coexisting atelectasis.

3. There was evidence of the passage of blood through the pulmonary vascular bed without exposure to alveolar air. This pulmonary shunt was probably due to continued perfusion of recently collapsed pulmonary tissue and accounted for most of the anoxemia found.

4. The postoperative increase in metabolic rate was accompanied by a proportionately greater increase in respiratory minute volume. An increase in the volume of dead space ventilated per minute accounted for most of the rise in the respiratory minute volume while the volume of alveolar ventilation increased only slightly. A fall in the respiratory quotient following decreased nutrition on the day of surgery explains the lack of a proportionate increase in the carbon dioxide eliminated in the presence of an elevated metabolic rate.

5. The nature of the stress thrown upon the organism by thoracoplasty may be summed up as follows: A chest whose expansion is limited by pain following surgical manipulation is required to maintain a slightly greater alveolar ventilation than it did preoperatively. This demand is met by an increased respiratory rate.

The authors are indebted to Miss Shirley Lundgren and Mr. John Ognanovich for valuable technical assistance.

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CIRCULATORY AND RESPIRATORY EFFECTS OF CONTINUOUS VERSUS PHASIC LUNG INFLATION IN OPEN-CHEST DOGS

Description of an Apparatus for the Application of Positive Pressure During Inspiration Only

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THIS study was undertaken to evaluate the degree of circulatory and respiratory disturbance which may be produced by continuous lung inflation as conventionally employed during thoracic surgery, and the extent to which such adverse effects may be diminished by a return to atmospheric pressure in the trachea during expiration.

A device which applies positive pressure during inspiration only was designed and built by one of us (A. M. C.). Its purpose is to minimize the physiologic disturbance inherent in positive pressure respiration, rather than to improve surgical convenience by controlling or suspending spontaneous respiratory movement. The apparatus not only removes the impediment to expiration, but allows the subject to initiate inspiration, so that the imposition of an arbitrary pattern of respiration is avoided.

METHODS

Healthy dogs premedicated with morphine sulphate (15 mg. intramuscularly), were anesthetized with ether until the lid reflex was lost. An effort was made to maintain this level of anesthesia throughout the experimental period. A tracheal cannula was placed, and a standard closed system established, permitting rebreathing through a freshly filled soda-lime cannister. The breathing bag was replaced by a cylindrical rubber bellows of several liters' capacity enclosed in an airtight glass cylinder. A vent in the cylinder maintained atmospheric pressure about the bellows, or else served to apply positive respiratory pressure. Tidal exchange was indicated by the excursion of the shadow of a wire mounted horizontally on the exterior of the glass cylinder projected to a vertical scale at the bottom of the bellows. Oxygen consumption was measured by noting the time required for the bellows to return to the null point after the introduction of 300 c.c. of oxygen by means of a water displacement apparatus. Dry gas volumes were corrected to standard conditions of temperature and pressure. Duplicate determination of oxygen consumption agreed within 3 per cent.

Mean respiratory pressures were indicated by a water manometer attached to a side-arm of the tracheal cannula. One femoral artery was exposed for blood samples, and the other was cannulated and attached to a mercury manometer. A catheter was introduced into the right auricle via the left internal jugular vein (position checked at end of experiment) for pressure measurements and mixed venous blood samples. Central (right auricular) and peripheral (femoral) venous pressures were measured by saline manometers, the zero point being set at the estimated level of the auricle.

Cardiac output was determined from oxygen consumption and arteriovenous blood oxygen difference by the Fick principle, and is expressed as "cardiac index" (L./min./m^2).

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An abstract of this paper was delivered before the American College of Surgeons in Chicago, October, 1949.

Received for publication Oct. 29, 1951.

Surface area was calculated from the weight.[1] Blood oxygen and CO_2 content were determined in duplicate by the manometric method of Van Slyke and Neill.[2]* Arterial oxygen saturation was calculated after determining oxygen capacity by equilibration in a tonometer or in a few instances by calculation from hemoglobin concentration (Evelyn colorimeter). The hematocrit of each samp'e was measured. Arterial blood pH was determined by the Beckman potentiometer, using an enclosed glass electrode. The CO_2 pressure and buffer base concentration were obtained from the nomogram of Singer and Hastings.[5] Electrocardiograms were taken from standard limb leads using needle electrodes.

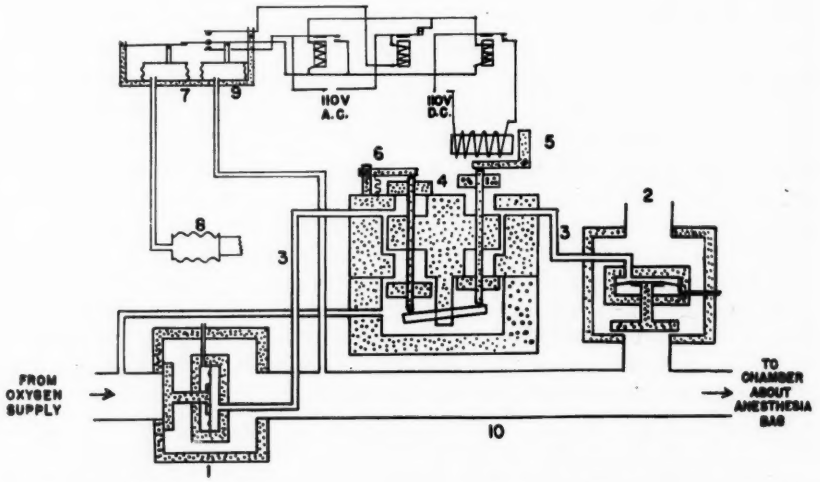

Fig. 1.—Schematic diagram of apparatus for inspiratory positive pressure respiration: the apparatus is designed to compress an enclosed anesthesia bag or bellows during inspiration and to allow a rapid return to atmospheric intratracheal pressure during each expiration. The compressing force is supplied by oxygen from a cylinder, the supply pressure having been reduced to a peak of 20 cm. water by a standard reduction valve and a type A-16 AAF spring-loaded demand regulator (not shown in the figure). Oxygen from this source provides the mechanical force required to open the inflow valve (*1*) and to close the exhaust valve (*2*) during inspiration. The gas pressure is applied to one or the other valve diaphragm (larger than the effective areas of the valve faces) by one of a pair of small ducts (*3*) leading from a set of reciprocal pilot valves (*4*). The pilot valves are brought to the inspiratory position by the action of an electromagnet (*5*), and are returned to the expiratory position by a spring (*6*). The magnet (direct current) is activated by a relay and holding circuit set in motion by the contraction of a metal bellows (*7*), which shortens as the rubber bellows (*8*) about the rib cage elongates at the start of inspiration. Expiration is initiated at the predetermined peak pressure by the elongation of a second metal bellows (*9*) in continuity with the main respiratory conduit (*10*). The electrical contact established by this second bellows (*9*) in its lengthened state breaks the circuit which has been supplying the electromagnet during inspiration, and allows the pilot valves to reverse to the expiratory position, so that the inflow valve (*1*) is closed by oxygen pressure, and the exhaust valve (*2*) is opened by the intracheal pressure, assisted by a spring (not shown) within the valve. The anesthesia bag then remains at atmospheric pressure until the next inspiration is begun.

The dogs were loosely held supine, and in each a chest wall incision was made which exposed the intercostal muscles along the full length of the right sixth interspace. Blood loss was held to a minimum. Initial observations were made one hour later. This preparation, i.e., normal respiration with closed chest, was studied as a control. In two other groups of dogs, the thoracotomy was completed and the wound edges were retracted to open the thorax widely. In Group CPP (continuous positive pressure) the vent in the glass cylinder

*The method of blood gas analysis was not modified to correct for the presence of ether in the blood because the magnitude of this error is small at this very light plane of ether anesthesia[3] and a satisfactory modification in technique[4] was not then available. It is considered that no error of interpretation was introduced since the blood ether concentration was kept nearly constant in each animal after a prolonged equilibration period, and was a factor common to all of the experimental groups.

enclosing the bellows was attached to a 2-liter rubber bag inflated to provide a mean intratracheal pressure of 10 to 13 cm. of water. This maintained a lung volume which just filled the chest in the expiratory position, with very little tidal exchange. In Group IPP (inspiratory positive pressure) the cylinder enclosing the bellows was attached to the device which applied positive pressure during inspiration (peak pressure 18 cm. H_2O; mean pressure 2.5 to 5.0 cm. H_2O), and allowed passive expiration against atmospheric pressure. The lung volume was observed to reach approximately one-half of full expansion during inspiration and to fall one-quarter expansion in expiration. Tidal volume was approximately 500 cubic centimeters.

All animals in the IPP and control groups tolerated the experimental procedure well, and a full set of observations was obtained at zero time (one hour after completion of preparations), at 1 to 1.5 hours, and again at 2 to 2.5 hours. Several CPP experiments had to be concluded within 1.5 hours because of marked fall in cardiac output and arterial pressure, whereas others could be carried for 2.5 hours.

Apparatus for Inspiratory Positive Pressure Respiration (Fig. 1).—The dog's chest was encircled by a rubber cuff connected to a hydron bellows so that inspiratory motion of the chest shortened the bellows and closed an electrical circuit which set the control valves into the inspiratory position and provided a free flow of oxygen at a maximal pressure of 20 cm. H_2O. Expiration was begun when the intratracheal pressure reached 18 cm. H_2O. At this pressure the expansion of a second hydron bellows interrupted the electrical circuit, and the return of the valves to the expiratory position permitted passive exhalation against atmospheric pressure (Fig. 2).

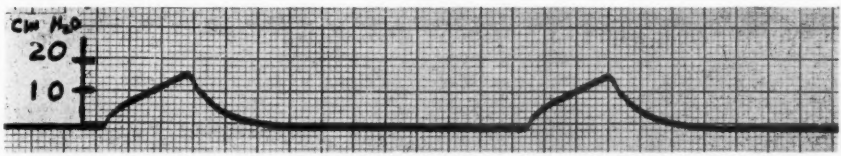

Fig. 2.—Sample intratracheal pressure curve during IPP respiration. An optical record taken from a membrane manometer attached to the intratracheal tube, converted to an electrical tracing by a pair of phototubes and inscribed by a direct-writing electrocardiograph. Calibrated by water manometer. Time interval between heavy vertical lines = 0.2 sec.

RESULTS

A. *Circulatory Effects* (Tables I, *A*, *B*, *C*).—The initial determinations of cardiac index ranged from 3 to 12 L./min./m², the mean values for each group being close to 7.5 L./min./m² (Fig. 3).* By the end of the subsequent 2 to 2.5 hours of observation the cardiac outputs of the control dogs were lower and more closely grouped in the normal range (average 3.6 L./min./m²). Arterial pressures remained normal and the total procedure, including the blood sampling, was tolerated well.

In four of the nine CPP (continuous positive pressure) dogs, the cardiac index declined below 2.4 L./min./m² within 1.5 hours, and in three others after 2 to 2.5 hours. The cardiac index of two CPP dogs remained normal throughout the experimental period. The average cardiac index at the end of the experiments was 2.9 L./min./m² (43 per cent of the starting values). A reduction in arterial pressure, accentuated by the withdrawal of blood samples,

*These high initial values may be attributed in part to the ether anesthesia.[6, 7]

TABLE I. A. CONTROL SERIES: DETERMINATIONS AT THE END OF THE PRELIMINARY EQUILIBRATION PERIOD (TIME ZERO) AND AT INTERVALS THEREAFTER

DOG NUMBER	TIME (HOURS)	CARDIAC INDEX (L./MIN./M²)	pH	pCO_2 (MM. HG)	BUFFER BASE (meq./L.)	ARTERIAL PRESSURE (mm. Hg)
1.	0	10.9	7.2	50	39	120
	1	13.9	7.20	44	37	120
	2	4.4	7.15	47	35	120
2.	0	12.4	7.20	63	42	100
	1	6.4	7.20	59	41	100
	2	3.1	7.20	53	40	100
4.	0	4.2	7.45	28	47	110
	1½	3.0	7.45	30	49	110
	2¼	3.8	7.40	33	46	110
5.	0	4.4	7.18	38	36	90
	1	7.1	7.10	48	34	100
	2¼	2.7	7.10	41	32	100
6.	0	5.7	7.20	40	36	120
	1¼	6.1	7.18	41	35	120
	2½	4.3	7.18	41	35	140
Averages	0	7.5	7.25	44	40	108
	1-1½	7.3	7.23	44	39	110
	2-2½	3.6	7.26	43	38	114

TABLE I, B. CPP SERIES: OPEN-CHEST DOGS BREATHING OXYGEN AT A PRESSURE SUFFICIENT TO MAINTAIN FULL LUNG VOLUME THROUGHOUT THE RESPIRATORY CYCLE

	DOG NUMBER	TIME (HOURS)	CARDIAC INDEX (L./MIN./M²)	PH	PCO_2 (MM. HG.)	BUFFER BASE (MEQ./L.)	ARTERIAL PRESSURE (MM. HG.)
	1.	0†	7.8	7.25	45	42	100
		1	2.3	6.85	130+	25	75
	2.	0	6.8	7.25	56	46	100
		1½	1.9	6.75	200+	10	110
	3.	0	9.0	7.30	52	48	110
		2	4.9	6.96	120	37	110
	4.	0	4.3	7.50	38	54	90
		1	3.4	7.40	45	50	90
Full lung inflation (Average respiratory pressure 10 to 13 cm. H_2O)	5.	0	6.2	7.30	30	38	110
		2½	5.2	7.05	45	28	110
	6	0	6.0	7.35	37	45	110
		2	2.3	6.85	130+	25	110
	7.	0	12.0	7.40	38	48	110
		1½	1.6	7.10	63	36	70
	8.	0	6.8	7.15	58	37	100
		¼	5.0	7.02	91	36	100
		1½	3.9	6.90	118	31	100
		2½	2.2	6.80	140+	27	100
	11.	0	6.5	7.45	34	48	100
		¼	2.7	7.10	82	40	90
		1	2.2	7.00	88	34	80
		2	2.1	7.00	88	34	80
Averages	(9 dogs)	0	7.3	7.36	43	45	103
	(6 dogs)	1-1½	2.5	7.00	107+	31	87
	(5 dogs)*	2-2½	3.3	6.93	103+	30	102
Lung half inflated (Respiratory pressure 4 cm. H_2O)	9.	0	8.5	7.25	53	43	100
		1½	2.3	6.77	150+	23	180
	10.	0	9.2	7.35	36	44	100
		1½	(dying)	6.81	150+	24	180

*Four dogs did not tolerate CPP respiration beyond 1.5 hours.

†Time zero refers to the end of the preliminary equilibration period and the moment before open thoracotomy and positive pressure respiration were established.

accompanied the decline in cardiac output in some of the animals, but marked elevation in arterial pressure occurred when the CO_2 accumulation was very great (Experiments 9 and 10, Table I, B).

The IPP (inspiratory positive pressure) group (seven dogs) showed the least decline in cardiac output, to an average of 5.4 L./min./m², or 85 per cent of the initial indices. In only two did the cardiac index fall below 3 L./min./m², and in these two the arterial pressure also declined.

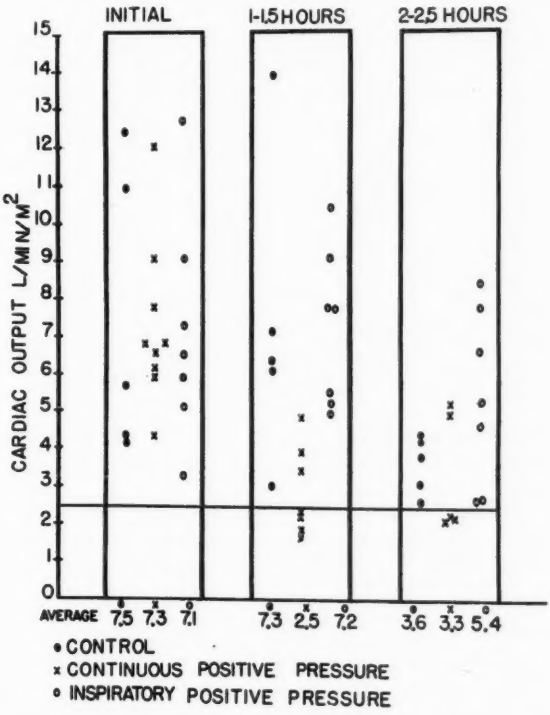

Fig. 3.—Cardiac output (L./min./m²) in the three groups of dogs at three phases of the experiment. The control dogs retained intact pleural sacs and breathed at atmospheric pressure. Open thoracotomy and positive pressure respiration was established in the other two groups immediately after the initial determinations of cardiac output.

The horizontal line at 2.4 L/min/m² represents the lower limit of cardiac output in normal dogs.[8, 9] Four dogs in the CPP group collapsed within 1.5 hours and are not represented in the third column. The two CPP dogs showing normal cardiac outputs at 2 to 2.5 hours are not represented in the 1 to 1.5 hours column. (CPP dogs 9 and 10, whose lungs were only half inflated, are not included in Figs. 3, 4, or 5.) All dogs in the control and IPP groups are represented in all three columns.

The initial femoral-auricular venous pressure gradient ranged from 1.0 to 6.5 cm. H_2O (average 4.0 cm. H_2O). Auricular and femoral venous pressure did not change during control observation. An elevation in both central and peripheral venous pressure occurred as soon as the chest was opened and positive respiratory pressure was applied (Table II). The new levels of venous pressure were reached within three to five minutes and did not change thereafter unless

TABLE I, C. IPP SERIES: OPEN-CHEST DOGS BREATHING OXYGEN INTERMITTENTLY SUPPLIED SO AS TO INFLATE THE LUNGS DURING INSPIRATION ONLY

DOG NUMBER	TIME (HOURS)	CARDIAC INDEX (L./MIN./M²)	PH	PCO_2 (MM. HG.)	BUFFER BASE (MEQ./L.)	ARTERIAL PRESSURE (MM. HG.)
4.	0†	7.3	7.35	39	44	100
	1	5.5	7.40	27	42	80-100
	2¼	2.7	7.40	22	38	80
5.	0	5.9	7.48	34	52	100
	1	7.7	7.35	48	49	100
	2	5.2	7.23	63	45	100
7.	0	3.3	7.50	32	52	110
	1	4.9	7.50	40	56	110
	2¼	6.6	7.50	40	56	110
8.	0	5.1	—	—	—	110
	1	9.1	7.50	27	49	100
	2	2.7	7.40	28	40	60
9.	0	6.5	7.20	56	42	100
	1	5.2	7.20	50	40	100
	2	4.5	7.20	52	41	100
10.	0	9.1	7.35	39	46	100
	1	7.7	7.40	39	50	100
	2	7.7	7.15	66	40	100
11.	0	12.7	7.30	36	41	100
	1	10.5	7.20	51	40	100
	2	8.4	7.20	62	43	100
Averages	0	7.1	7.36	39	46	103
	1	7.2	7.36	40	47	100
	2-2¼	5.4	7.29	47	44	93

†Time zero refers to the end of the preliminary equilibration period and the moment before open thoracotomy and positive pressure respiration were established.

the respiratory pressures were altered. In the CPP group, the mean increase in auricular pressure was 5 cm. H_2O, and in the femoral pressure 4 cm. H_2O. In experiments 9 and 10 in which the lungs were only partly inflated the largest

TABLE II. RELATIONSHIP OF VENOUS PRESSURE CHANGE TO INCREASE IN RESPIRATORY PRESSURE DURING CPP AND IPP RESPIRATION IN OPEN-CHEST DOGS

SERIES	DOG NUMBER	INCREASE IN RESPIRATORY PRESSURE (CM. H_2O)	INCREASE IN AURICULAR PRESSURE (CM. H_2O)	CHANGE IN FEMORAL-AURICULAR VENOUS PRESSURE GRADIENT (CM. H_2O)
	1.	10	5.0	−2.0
	2.	13	7.0	0
	3.	10	5.5	0
	4.	10	–	–
CPP	5.	10	6.0	−2.0
	6.	10	6.5	−1.5
	7.	10	5.0	−1.5
	8.	10	4.5	−0.5
	11.	11	4.0	−0.5
	Mean	**10**	**5.0**	**−1.0**
	9.	4	9.0	+0.5
(half-inflation)	10.	6	10.0	−3.5
	4.		4.0	0
	5.		4.0	−1.5
	7.		4.5	−3.0
IPP	8.	2.5 to 5.0	4.0	−1.0
	9.		1.5	+1.0
	10.		1.0	+1.0
	11.		2.0	+1.0
	Mean	**2.5 to 5.0**	**3.0**	**0**

increase in central venous pressure, 9 to 10 cm. water, was reached. These are not included in this average. In the IPP group, the mean increase of 3 cm. H_2O in central venous pressure was accompanied by an equal increase in femoral venous pressure. Changes in venous pressure gradient in individual dogs could not be correlated with changes in cardiac output.*

Study of the electrocardiograms in three leads disclosed no significant alteration in conduction, electrical activity or in the mechanism of the heartbeat.

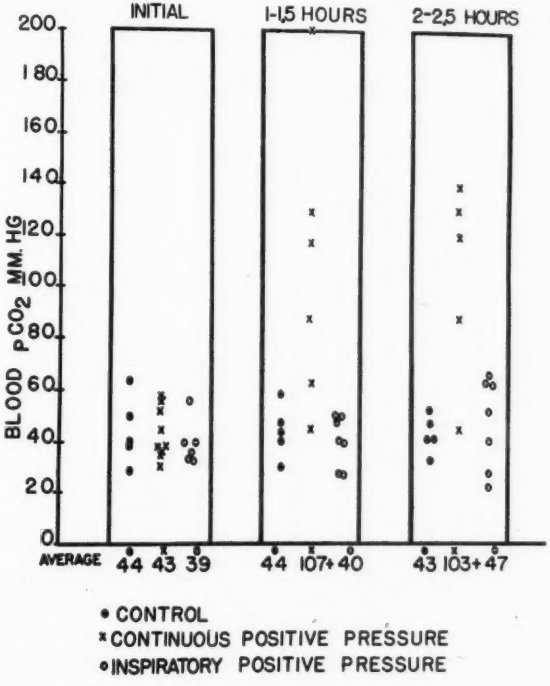

Fig. 4.—Arterial blood pCO_2 (mm. Hg) in the three groups of dogs at three phases of the experiment (as described in the legend of Fig. 3).

B. *Respiratory Effects* (Tables I, A, B, C).—Arterial oxygen saturation remained above 95 per cent in all dogs except for four in the CPP group in which the values lay between 87 per cent and 91 per cent.

Arterial pCO_2 (mean initial values 39 to 44 mm. Hg) remained unchanged in the control dogs, but rose to extreme values during CPP respiration (Fig. 4). The average arterial pCO_2 at the end of the period of CPP respiration was 107

*The pressure determined in any selected vein is but a sample of a "peripheral venous pressure," and cardiac output cannot be expected to be strictly proportional to changes in femoral vein—right auricular gradient. Furthermore, the pressures recorded are mean pressures, so that a zero gradient need not mean zero flow. Forward flow will continue in those phases in which the gradient is positive, while backflow during phases of negative gradient is prevented by the venous valves.

mm. Hg in the nine dogs maintained at full lung inflation, and was 150 mm. Hg in the two dogs whose lungs were half inflated. During IPP respiration, in contrast, there was little or no CO_2 accumulation.*

Acidosis of severe degree developed regularly during CPP respiration (Fig. 5). The arterial pH dropped from the mean initial value of 7.36 to a mean of 6.97 by the end of the period of CPP respiration. In one dog the pH dropped from 7.45 to 7.10 and the pCO_2 rose from 37 to 82 mm. Hg within fifteen minutes of CPP respiration. Another animal reached a pH of 6.75 and a pCO_2 above 200 mm. Hg within one and one-half hours. In most animals metabolic acidosis accompanied the respiratory acidosis which developed during CPP respiration (Fig. 6).† The average buffer base level declined from the control value of

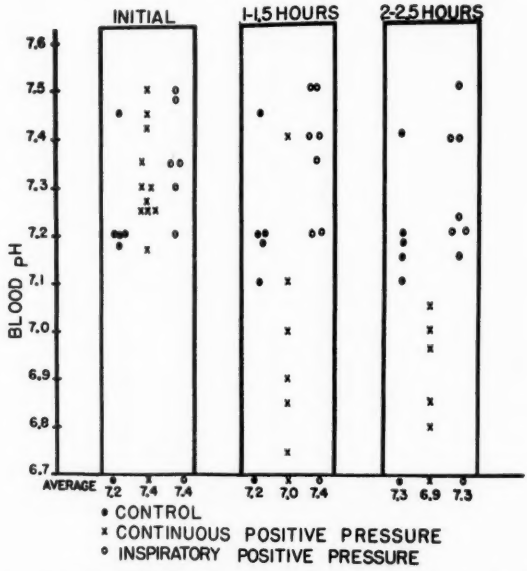

Fig. 5.—Arterial blood pH in the three groups of dogs at three phases of the experiment (as described in the legend of Fig. 3).

45 meq. per liter to 30 meq. per liter by the end of the experimental period. In no instance was there evidence of an increase in concentration of fixed base which might have been interpreted as a compensatory change in response to respiratory acidosis. The degree of decline in cardiac output could not be correlated with the degree of acid-base disturbance.

IPP respiration throughout the period of open-chest observation prevented any significant change in blood pH.

*The IPP instrument can produce marked overventilation and CO_2 depletion if not controlled by the dog's respiratory rhythm. In two discarded experiments an electrode attached to a nostril was used to initiate inspiration. This proved to be oversensitive to extraneous movement and overbreathing was produced, so that arterial pCO_2 fell below 11 mm. Hg. and the arterial pH rose to 7.6. The cardiac outputs of these two dogs remained normal.

†Mild metabolic acidosis was observed in a number of dogs at the time of the initial blood sample. Metabolic acidosis may be attributed to ether anesthesia in dogs,[11, 12] although apparently not in man.[13]

In three supplementary experiments an effort was made to correct the respiratory acidosis of CPP respiration by brief periods of repeated emptying and filling of the lungs, such as might be done manually by an anesthetist. In D1, the lungs were allowed to empty and were then reinflated; this cycle was repeated four times in immediate succession. Emptying proceeded so slowly that ventilation was further impaired rather than improved, and the acidosis which had been mild (pH 7.2, pCO_2 45 mm. Hg) was intensified (pH 7.05, pCO_2 70 mm. Hg). In D2, after an hour of CPP respiration, the pH was 7.0 and the pCO_2 106 mm. Hg. During a subsequent ten-minute period of IPP respiration, using the device herein described, these values remained unchanged. In D3, CPP respiration was interrupted and the lungs partially deflated and inflated twice every fifteen minutes. Nevertheless, severe respiratory acidosis developed within an hour (pH 6.92, pCO_2 116 mm. Hg).

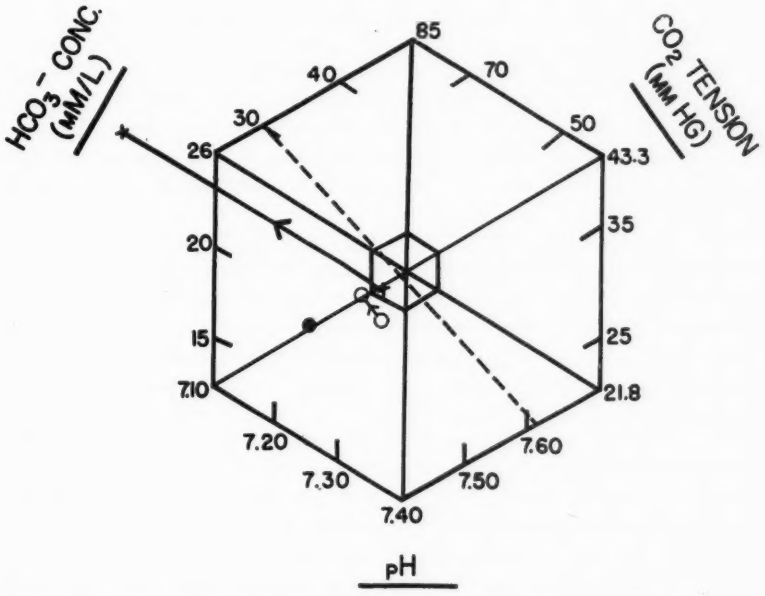

Fig. 6.—Triaxial diagram of pH, pCO_2, and bicarbonate concentration, adopted from Hastings and Steinhaus.[10] The broken line indicates the path of uncomplicated CO_2 increase. The solid circle represents the average of control values. No change occurred during control observation. The open circles indicate the average of values before and after IPP respiration. The change is small. The crosses indicated the values before and after CPP respiration. Marked acidosis and CO_2 accumulation are denoted. The deviation of the CPP path to the left, with reference to the broken line of uncomplicated CO_2 accumulation, indicates the metabolic component.

DISCUSSION

In intact animals undergoing positive pressure respiration, reduced ventilation is less critical than the interference with circulation consequent to elevation of intrapulmonary and intrapleural pressure.[14-30] In the open-chest animal, however, the rise in intrapleural pressure is limited, so that the decline in

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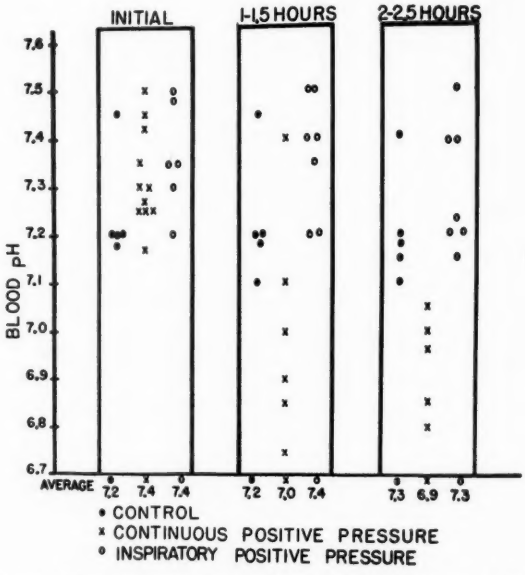

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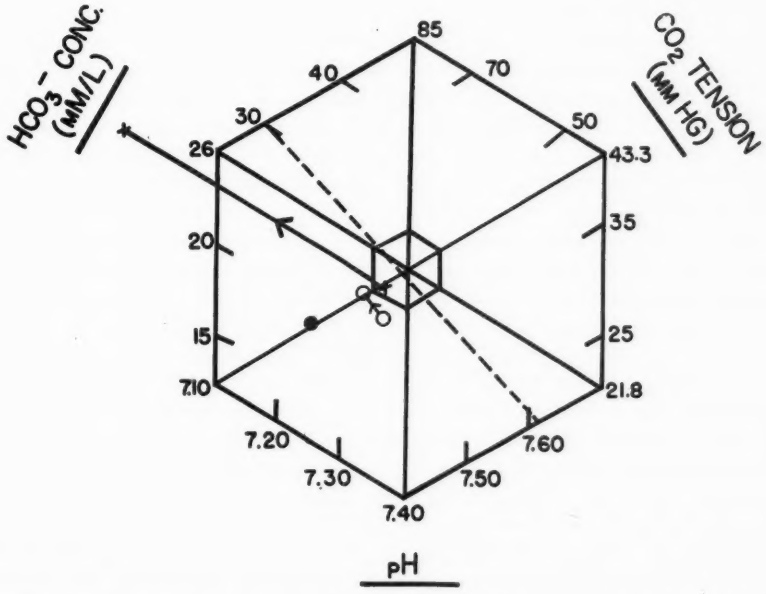

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DISCUSSION

In intact animals undergoing positive pressure respiration, reduced ventilation is less critical than the interference with circulation consequent to elevation of intrapulmonary and intrapleural pressure.[14-30] In the open-chest animal, however, the rise in intrapleural pressure is limited, so that the decline in

ventilation is the most obvious adverse effect of continuously maintained positive respiratory pressure. But cardiac output also falls and to a greater degree than might be expected on the basis of the intrapleural pressure change alone.[31-33] Since the decrease in cardiac output is greater during CPP (continuous positive pressure) respiration than during IPP (inspiratory positive pressure) respiration, the level of intrapleural pressure, which is presumably the same in both (open-chest), is not the only controlling factor. Hence the mean level of intrapulmonary pressure also influences cardiac output in these circumstances. An increase in intrapulmonary vascular resistance[16, 34] is suggested by the greater increase in right auricular pressure during CPP respiration.*

The CPP animals might have been expected to have the higher cardiac outputs because of the exaggerated respiratory effort accompanying CO_2 accumulation, and because acidosis as well as CO_2 accumulation are said to increase cardiac output.[36, 37] The observed contrary effect is not attributable to a greater degree of tachycardia[38] nor was there evidence of plasma volume loss[39] or inadequacy of "effective venous pressure."† Since respiratory motion produces blood flow even in the absence of heartbeat,[42] its absence during CPP respiration may partially account for poorer circulatory function than during IPP respiration. Except for this pumping action, the favorable effect of IPP respiration upon the circulation should probably be attributed to the lower mean intrapulmonary pressure, rather than to the intermittency of the positive pressure application.

The arterial pressures observed during CPP experiments were presumably the resultant of the opposing effects of high CO_2 tension[43] and reduced cardiac output. Poor tolerance of blood sampling was manifested by animals with low cardiac output, whether the arterial pressure was normal or low.

Open-chest animals with lungs maintained in continuous inflation (CPP) have so little tidal exchange that the elimination of CO_2 is inadequate. Severe respiratory acidosis results, but the cause of the accompanying metabolic acidosis is obscure. The lowered cardiac output may produce tissue anoxia and reduced renal function, with a resulting accumulation of lactate, pyruvate, phosphate, and sulfate ions, but the reduction in buffer base is greater than the observed degree of peripheral vascular disturbance can account for.[44]

Allowing the lung to collapse completely and then refilling it with oxygen is not an effective technique of clearing respiratory acidosis and may make it worse. Periodic partial emptying and reinflation of the lung at intervals of fifteen to thirty minutes, such as is commonly done by the anesthetist during open-chest surgery, does not prevent CO_2 accumulation. Even a return to effective ventilation does not correct respiratory acidosis as rapidly as it develops. This may be because intracellular changes which have occurred during the period of CO_2 accumulation are not immediately responsive to reduction in alveolar CO_2 tension. A similar delay in improvement, indeed an initial in-

*However, partial collapse of the lungs also produces high pulmonary vascular resistance.[35] The highest auricular pressure in the current experiments were found in the two dogs (CPP 9 and 10) whose lungs were allowed to remain half collapsed at low respiratory pressure.

†The "effective venous pressure" is defined[40] as the pressure available for filling the relaxed ventricle and is the gradient from auricular to extracardiac (intrapleural) pressure[41] This was above the critical limit for right ventricular output (5 cm. H_2O)[40] in all experiments.

crease in acidosis, has been observed during resuscitation following experimental asphyxia.[45] Phasic interruption of the applied respiratory pressure to permit free exhalation (IPP) maintains effective ventilation throughout the open-chest period.

The human subject appears to withstand prolonged open-chest surgery under a form of positive pressure respiration which is poorly tolerated by the dog. With a unilateral thoracotomy he ventilates somewhat better than the dog, in which the flimsiness of the anterior mediastinum prevents any expansion of the contralateral lung. This advantage may[46] or may not[47] be reduced in the lateral position, in which expansion of the intact hemithorax is limited by the patient's weight. In the prone position a congested or relatively inelastic human lung requires little or no positive respiratory pressure to maintain partial expansion. This may reduce the disturbance of circulatory dynamics but it should not improve ventilation. It has recently been shown that a severe degree of respiratory acidosis may indeed develop within two hours of human open-chest surgery[48, 49] and that this is accompanied by an unexplained metabolic acidosis. Respiratory acidosis is less likely to occur when the patient is supine, or suspended by the shoulders and pelvis in the prone position so that the thorax bears no weight, than in lateral or simple prone position.[50, 51] The level of anesthesia also plays a part, for respiratory acidosis may be observed during deep narcosis for operations other than thoracotomy.[52] The transfusion therapy routinely applied to compensate for blood loss and surgical trauma probably also tends to correct the decline in circulation resulting from positive pressure respiration, but the degree of circulatory inadequacy which remains is not known. Elevation in arterial pressure produced by the increase in CO_2 tension may give a falsely favorable impression of the state of the circulation. The ordinarily transitory rise in pulmonary arterial pressure and fall in cardiac output accompanying ligation of a major pulmonary artery[53, 54] may be accentuated by positive pressure respiration. The human heart is less able to tolerate severe acidosis than is the dog's heart, as judged by electrocardiographic evidence.[55] Our studies agree with those of others in not finding ECG changes in the CPP dogs,[55, 56] but evidence of myocardial damage has been found to accompany severe CO_2 acidosis in well-oxygenated patients.[55] Respiratory acidosis during cardiac surgery has been reported to increase susceptibility to acute arrhythmias.[57] CO_2 accumulation produces a marked rise in cerebrospinal fluid pressure.[58] It is apparent, then, that continuous lung inflation is not entirely satisfactory for clinical intrathoracic surgery and may account for a number of adverse reactions.

Mechanically imposed ventilation is applied during open-chest surgery in a number of clinics.[59-63] Manual compression and release of the breathing bag throughout the procedure may prevent respiratory acidosis.[50] Controlled respiration, whether accomplished manually or by automatic device, in the absence of continuous and precise monitoring of arterial blood CO_2 pressure, pH and oxygen tension is unlikely to be as responsive to the patient's needs as spontaneous respiration. The apparatus herein described is under the control of the

subject's respiratory center. The data presented indicate, for dogs at least, that the performance of the respiratory center remains satisfactory under light ether anesthesia.

SUMMARY

Continuous inflation of the lungs of open-chest dogs for a period of two hours or less is usually accompanied by a substantial decline in cardiac output and the development of respiratory acidosis of severe degree. These adverse effects may be minimized or avoided by interrupting the positive respiratory pressure to permit passive exhalation against atmospheric pressure. A device is described which applies positive respiratory pressure during inspiration only, and which is controlled by the animal's own respiratory rhythm.

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SUCCESSFUL REMOVAL OF TUMOR OF THE HEART

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TUMORS of the heart, both primary and metastatic, are comparatively rare. According to an excellent review of the subject by Prichard[1] in January, 1951, approximately 500 metastatic tumors of the heart and 415 primary ones have been reported in the literature. Of the primary tumors, 112 were sarcomas, and 303 were benign lesions. According to Prichard,[1] who quotes from Martin's report of 1929, only twenty-one lipomas are included in the series of benign primary cardiac tumor cases. A review of Martin's[2] publication, however, reveals that only one incidence of lipoma of the heart was presented therein by the author. Twenty additional cases of lipoma were referred to in the literature, but were not confirmed by bibliographical references.

A second case report of lipoma of the heart, in a 7-month-old baby girl, was described by Brewis[3] in 1905. At necropsy, a "fatty tumor" was encountered arising from the interventricular septum. The tricuspid valve was obstructed by the portion of tumor which protruded into the right ventricle. As accurately as could be determined, only two proved cases of lipoma of the heart have been reported. No lipoma of the heart has been diagnosed ante mortem, nor has one been successfully removed by operation. Of the 303 benign tumors of the heart reported in the literature, only five were diagnosed before death of the patient.

Successful surgical removal of a cardiac tumor is exceedingly rare. Only one instance has been recorded by Beck[4] in 1942. Two patients, according to Beck, were operated upon successfully, but in only one instance was a true tumor removed. The first operation was performed in two stages, in 1934 and 1936, upon a white man 22 years of age. An intrapericardial teratoma, which arose from the right ventricle and was intimately adherent to the aorta, pulmonary artery, vena cava, and right inferior pulmonary vein, was successfully removed. The second operation was performed by Beck in 1940 upon a 39-year-old white man. A calcified cyst, without any pathologic evidence of true neoplastic formation, was removed from the left ventricular wall. Both patients survived and did well after surgery. No other reports of the successful removal of a cardiac tumor have been encountered in this author's review of the literature.

In 1941-1942, Gebauer[5] attempted, in three stages, operative removal of a teratoma and cyst from the heart of a 12-year-old girl. Unfortunately, the patient expired on the operating table because of uncontrollable hemorrhage from a perforation in the aorta, produced inadvertently during the application of a clamp across the tumor pedicle.

Received for publication Oct. 17, 1951.
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The following case report is presented because of the extreme rarity of the lesion involved, the unusually large size of the tumor (3½ pounds), and because it is the first recorded instance of successful surgical removal of a primary lipoma of the heart.

CASE REPORT*

C. D., A 44-year-old white man, was first seen by Dr. Henry F. Kulle of Cincinnati, because of bleeding from the rectum, easy fatigability, and vague discomfort over the anterior chest. There was no history of cardiac decompensation, cough, hemoptysis, or remote pulmonary disease. Fluoroscopic examination of the patient's chest by Dr. Kulle showed the cardiac silhouette to be greatly enlarged in all directions.

The patient was admitted for further study to the Good Samaritan Hospital of Cincinnati on July 10, 1951.

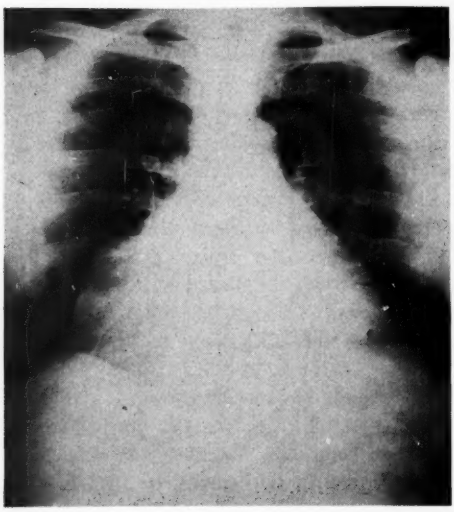

Fig. 1.

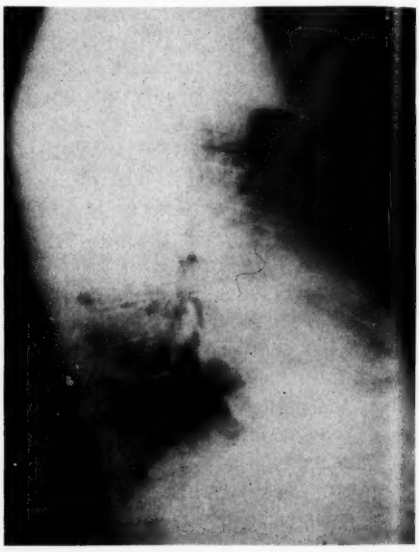

Fig. 2.

Figs. 1 and 2.—Posteroanterior and lateral projection roentgenograms showing enlargement of the cardiac silhouette.

Physical Examination.—The patient was a well-developed, moderately obese white man, who did not appear to be in any acute or chronic distress. The temperature was normal, blood pressure was 110/80, and the pulse 85 per minute. No arrhythmias nor paradoxical pulses were noted. There was no distention of the neck or thoracic veins, which would suggest any elevation in the venous pressure. The arm veins emptied rapidly on elevation. The lungs were entirely clear to percussion and auscultation, and the breath sounds were somewhat distant. No adventitious sounds were audible. The heart sounds were of good quality, but were somewhat muffled and distant. The apex impulse was difficult to locate by palpation. There seemed to be very marked enlargement of the cardiac diameter by percussion. The rest of the physical examination revealed entirely normal findings.

Roentgenograms.—Preliminary roentgenograms of the chest, posteroanterior and right lateral projections (Figs. 1 and 2), showed enlargement of the cardiac silhouette in all dimensions, particularly to the right of the midline. Barium studies of the entire gastroenteric tract revealed no evidence of organic lesions.

*The author is indebted to Dr. Henry F. Kulle for permission to publish this case report.

An electrocardiogram made on July 12, 1951, revealed essentially normal findings, except for prolongation of the auriculoventricular conduction time to a high normal, and moderate left axis deviation. The tracing was thought to be within normal limits.

Laboratory examinations, including urinalysis, complete blood cell counts, and hemoglobin determinations, revealed normal findings.

An angiocardiogram (Fig. 3) performed on July 11, 1951, with 70 per cent diodrast, showed excellent filling of the inflow tract, including the superior vena cava, right auricle, and a portion of the right ventricle. Some dye was also seen in the left side of the heart. The greater portion of the density seen on the posteroanterior projection roentgenogram of the chest appeared to be due to an extracardiac tumor or cyst, which was not attached to the heart. A provisional diagnosis of pericardial coelomic cyst was made. Twenty-four hours preceding operation, the patient received S. R. penicillin, 400,000 units, and one-half gram of streptomycin hypodermically, every twelve hours.

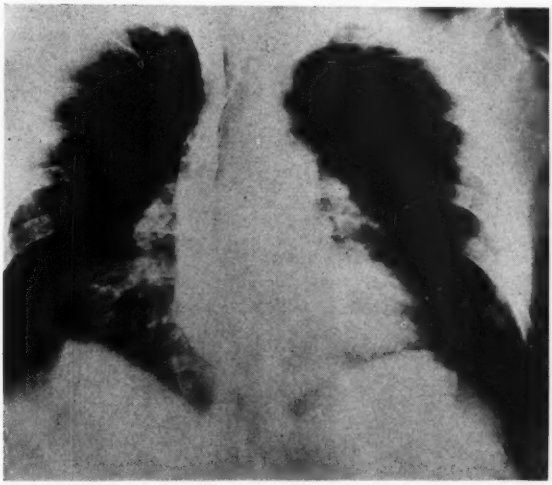

Fig. 3.—Angiocardiogram showing excellent filling of the inflow tract and the extracardiac location of the tumor.

Operation.—July 14, 1951, at Good Samaritan Hospital. With the patient in the left lateral prone position and under endotracheal gas, oxygen, and ether anesthesia, a curved periscapular incision was made over the right side of the thorax. The sixth rib was subperiosteally resected in its entirety, and the pleural cavity was entered through the rib bed. The lower and middle lobes of the right lung were adherent to the pericardium and diaphragm by numerous broad adhesions, which were divided by sharp dissection. The pericardium was extremely thin, transparent, and distended to approximately ten times normal dimensions, so that it bulged into the right chest. A normal-sized phrenic nerve coursed in the usual anatomic position, over the outside of the pericardial sac. Through the thin, transparent pericardial wall, numerous coils of yellowish gray tissue could be seen, which on first inspection looked not too much unlike loops of intestine. As soon as it was determined that manipulation of the heart would be necessary, the patient was given 20 c.c. of 1 per cent procaine by intravenous drip, to decrease the sensitivity of the heart, and to prevent arrhythmias during manipulation. A longitudinal incision was made in the pericardial sac anterior to the phrenic nerve, and the sac was opened widely. Almost immediately, numerous large loops and coils of adipose tissue bulged into the right chest. The entire tumor was completely delivered down to a common stalk, which was attached to the anterior surface of the left ventricle, over a circular area which measured approximately 4 by 4 cm. in diameter. At

this point in the operation, it was obvious that the lesion was a massive lipoma, arranged in loops and coils, and arising by a common pedicle from the left ventricular wall. The point of attachment of the lipoma to the ventricular wall was separated without too much difficulty, by alternate sharp and blunt dissection. As nearly as could be determined, the tumor arose from the superficial layer of the myocardium beneath the epicardium, and penetrated the myocardium for a distance of several millimeters. Following the separation of the tumor pedicle, there was a moderate amount of brisk bleeding from the surface of the heart. Persistent bleeding from one moderate-sized vessel was controlled with warm moist compresses, rather than suture ligature, because of the proximity of the bleeder to the coronary artery. Approximately 150 c.c. of blood accumulated, as a result of the bleeding from the amputation site. During the course of the operation, the chamber of the heart was not entered. Examination of the interior of the tremendously dilated pericardial sac showed it to be in continuity and without any connection with the abdomen or contralateral hemithorax. Grossly, the heart appeared to be entirely normal. The superior vena cava was not dilated and showed no evidence of previous obstruction.

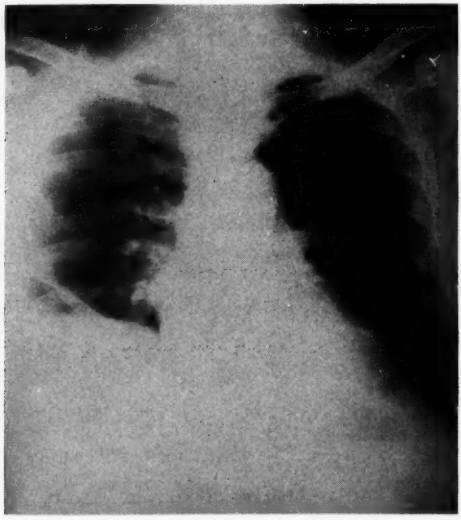

Fig. 4.—Posteroanterior projection roentgenogram of the chest showing return of the cardiac silhouette to normal size following removal of the tumor.

It was the author's impression that the easy distensibility of the pericardial sac and the slow growth of the tumor presumably accounted for the absence of venous obstruction and elevated venous pressure. The interior of the pericardial cavity was irrigated thoroughly with saline solution, and was then aspirated dry, following which procedure the incision in the pericardium was closed with interrupted sutures of medium silk. A small window, three to four centimeters in diameter, was allowed to remain in the inferior portion of the pericardial incision, to permit drainage of the pericardial cavity into the right chest. At this point in the operation it was noted that the right phrenic nerve had been inadvertently crushed by a clamp, which had been placed along the margin of the pericardial incision for retraction. This resulted in partial paralysis of the right, diaphragmatic leaf. The right chest was lavaged with saline solution, and was then aspirated dry, following which procedure the right lung was re-expanded by positive pressure and manipulation. The chest wound was closed by interrupted medium silk sutures, which were applied to the periosteal rib bed, intercostal muscles, chest wall, muscles, fascia, and skin. A dry sterile dressing was then applied, and the patient was returned to his room in good condition.

Following the operation the patient had a very uneventful course. He was ambulatory by his second postoperative day, and was discharged from the hospital, with the wound healed, on the ninth postoperative day.

When seen in a follow-up examination, two and one-half months after the operation, the patient was entirely without complaints, except for some discomfort over the anterior portion of the chest wound. There was no complaint of dyspnea, palpitation, or precordial discomfort. A roentgenogram of the chest made two months following the operation (Fig. 4) showed the cardiac silhouette to be approximately normal in size. The right lung was completely re-expanded, and there was no evidence of effusion. The right diaphragm remained partially elevated, as a result of the phrenic nerve crush at the time of operation.

Pathology.—Grossly, the tumor consisted of numerous coils and loops of fat tissue, which arose from a common stalk or pedicle (Fig. 5). The fresh specimen weighed 3.5 pounds. Microscopic examination revealed typical fat cells, with irregular limits (Fig. 6). There was no evidence of malignant degeneration in any portion of the tumor.

Diagnosis.—Lipoma.

Fig. 5.—Photograph of gross specimen. Arrow indicates point of attachment of tumor to the heart.

DISCUSSION

Primary lipoma of the heart is a pathologic rarity. In a review of the literature only two authenticated cases are recorded. There seems to be no record in the past of successful surgical removal of a cardiac lipoma. A study of the case reports indicates that this is the largest primary tumor of the heart ever reported, including all varieties.

A more accurate preoperative diagnosis could possibly have been made by utilizing a diagnostic pneumopericardium, as suggested by Leubschitz, Lundsteen, and Forchhammer.[6] The use of a preoperative diagnostic pneumopericardium was discussed but finally decided against, because of the history and angiocardiographic findings, which suggested a pericardial coelomic cyst. In retrospect, an artificially induced pneumopericardium before the operation would undoubtedly have settled the issue.

Not encountered in the case reported were certain classical symptoms and signs of tumor of the heart, such as decompensation developing without demonstrable cause and proving absolutely refractory to treatment, arrhythmias, failure of conduction as demonstrated on the electrocardiogram, and hemorrhagic pericardial effusion.

The prognosis in cardiac tumors is bad, as a rule, even in the case of benign tumors. There are, however, cases on record, in which symptoms of intracardiac myxomata were present for many years, as in the example reported by Strouse[7] in 1938. The duration of the disease in this instance was forty-three years.

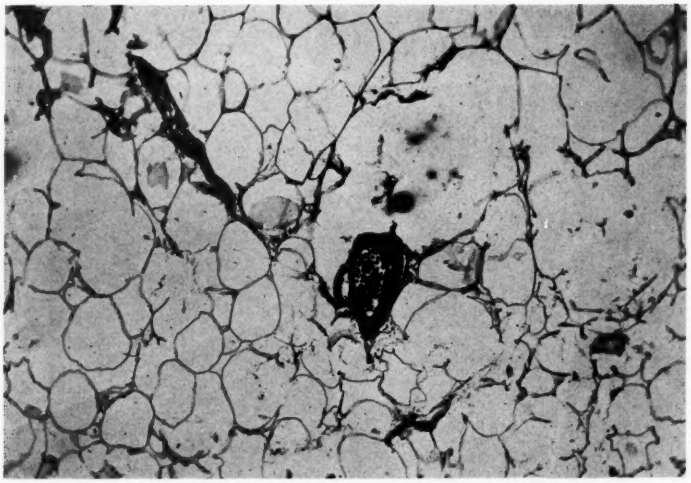

Fig. 6.—Microphotograph of the removed tumor. Typical fat cells are apparent without evidence of malignant change.

There are several reports, also, of the incidental finding at necropsy, of benign tumor of the heart, which had been, clinically, completely or almost completely silent. Such cases were reported by Arneson[8] in 1925 and by Guldberg[9] in 1928. In both instances, the tumor was a fibroma in the atrium. As a rule, malignant cardiac tumors run a comparatively brief course, and the outcome is invariably fatal.

The general feeling of the futility of treating primary tumors of the heart is not borne out by experiences during recent years, with the roentgenologic and operative treatment of these lesions. In 1937, Popp[10] reported a case of sarcoma of the right atrium, in which the symptoms improved, following x-ray radiation therapy instituted for the purpose of diagnosis. In 1940, Shelburne and Aronson[11] described a case of metastatic myeloblastoma in the heart, which was diagnosed by heart block and pericardial exudate, containing tumor cells. Following x-ray radiation therapy, the roentgenologic picture returned to normal, the heart block became partial on the electrocardiographic examination, and the patient was symptom free for many months, until metastases appeared in other

organs. In 1940, Hsiung, Fzutu, Hsiehn and Lien, as quoted by Leubschitz, Lundsteen and Forchhammer,[6] also described the good effects of roentgen treatment in a metastatic cardiac tumor, which had arisen from a primary reticular sarcoma of the nose. In this particular instance, the roentgen picture, which had shown an enlarged heart shadow with kymographic signs of exudate, became normal following the roentgen treatment. The extremely low mortality rate attending exploratory thoracotomy, the availability of the modern antibiotics and the intriguing possibilities of intracardiac surgery, which might become possible following the clinical use of extracorporeal heart and lung machines now on the horizon, should make possible, in the near future, the surgical removal of a very large number of benign tumors, such as myxomata.

Myxomas comprise the bulk (50 per cent) of the benign tumors commonly encountered in the heart. They are most usually pedunculated, and they arise from the endocardial surfaces of the auricles, so that they would be particularly amenable to surgical removal.

SUMMARY AND CONCLUSIONS

1. A successful surgical removal of a primary lipoma of the heart is reported. As nearly as can be determined, this is the first report of a successful removal of a primary lipoma of the heart and the second instance of a successful removal of a tumor of the heart, including all varieties.

2. The tumor in the case presented is the largest primary one, malignant or benign, which has been found reported in the literature to date.

3. Primary lipoma of the heart is a pathologic rarity, only two proved cases having been reported previously.

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SURGICAL TREATMENT OF EPIPHRENIC DIVERTICULUM OF THE ESOPHAGUS

Review of Literature and Report of Case Treated by Resection and Esophagogastrostomy

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DIVERTICULA of the lower portion of the esophagus are relatively uncommon, whereas those located in the terminal portion just above the diaphragm, termed supradiaphragmatic or epiphrenic diverticula,[38] are even less frequently encountered. In a review of the literature up to 1933, Granet was able to collect forty-nine cases, only thirty-one of which he accepted as proved cases of epiphrenic diverticula. Vinson, who reviewed the records of the Mayo Clinic up to 1934, discovered forty-two cases of diverticula of the thoracic esophagus, twenty-four of which were located in the lower third; less than half of these patients were considered to have symptoms that could be definitely associated with the diverticulum, a fact which indicates another reason why they are infrequently encountered as clinical problems. In an analysis of 1,600 patients admitted to the Outpatient Department of Massachusetts General Hospital because of dysphagia, Macmillan found less than one per cent due to diverticula of the thoracic esophagus. Further evidence of the rarity of this form of esophageal diverticulum is found in its relative incidence in comparison with the pharyngoesophageal type. Thus, Harrington encountered only eight cases of the epiphrenic type during the same period that he treated surgically 216 patients with pharyngoesophageal diverticula, and Lahey[26] reported a somewhat similar experience with six cases of the former to 211 of the latter type.

The paucity of the literature on the surgical management of epiphrenic diverticula of the esophagus is a further reflection of their infrequent occurrence, at least as clinical problems. Even as late as 1949 Poppe and Berg, in a report of a case of a patient successfully treated by diverticulogastric anastomosis, stated that in their review of the literature they had been able to find no previous report of the successful application of this method of treatment and only one case of successful resection. Although a more careful review of the literature has disclosed a number of previously recorded cases in which both of these methods of treatment had been successfully applied, there is nonetheless a dearth of reports on the surgical management of this type of esophageal diverticulum. For these reasons, and because no report has been found of the clinical application of the technical procedure used in the case to be described, a brief review of the surgical considerations of epiphrenic diverticula of the esophagus seems desirable.

These diverticula are generally considered to be of the pulsion type resulting from herniation of the mucous membrane through the muscular wall, probably in an area of congenital weakness, although the etiology is still ob-

Received for publication Oct. 22, 1951.
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scure. The opening of the pouch is usually located in the posterior wall, more often directed toward the right than the left side, about 5 to 10 cm. above the cardia. The wall is composed of mucosa, submucosa, muscularis mucosa, and an outer fibrous layer of varying density. Infiltration of the wall of the sac with inflammatory cells is common, and the mucosa may become ulcerated. As the sacculation increases in size, it tends to assume a more dependent position in relation to the esophagus with distortion, angulation, and partial obstruction of the esophagus distal to the opening of the pouch. Accordingly, food passing down the esophagus tends to be shunted into the pouch. Cardiospasm has been considered a frequently associated factor and even of etiologic significance by some,[7, 19, 33, 35, 37, 44, 49] but we, like others,[2, 20, 21, 34] doubt this relationship, since diverticula are rarely found in patients with cardiospasm or achalasia. Probably the commonest complications are inflammation of the wall of the diverticulum, ulceration, and associated esophagitis, but perforation, hemorrhage, and malignancy have occasionally been encountered. Stricture of the esophagus distal to the diverticulum, which in the case to be described was largely responsible for the severity of symptoms, is not an unlikely complication in view of the progressive distortion and obstruction of the esophagus by the diverticula and the consequent prolonged esophagitis and spreading chronic inflammatory reaction.

Until rather recently most authors writing on this subject advocated conservative treatment because most patients had few or no symptoms, and operation was associated with considerable risk. It is now generally agreed, however, that surgical treatment is the method of choice and should be considered in all esophageal diverticula-producing symptoms. The surgical procedures that have been employed in the treatment of epiphrenic diverticula may be classified into four categories: (1) anastomosis between the sac and the stomach, (2) extirpation of the sac, (3) diverticulopexy, and (4) inversion of the sac. Historically, anastomosis between the diverticulum and the stomach was the procedure advocated, having been proposed in 1908 by Lotheissen, although Gosset, in 1903, had suggested this procedure for strictures of the cardia and had demonstrated its feasibility in cadavers and dogs, using a transpleural approach. In a subsequent publication Lotheissen[29] referred to a case in which this procedure was done using an abdominal transdiaphragmatic approach. Other German surgeons[15, 17, 22, 41] have employed this type of operation with apparently good results, while Henschen has been credited with the report of the first successful case in which this operation was done from above the diaphragm. Sauerbruch[41] advocated performance of the procedure in two stages, the first consisting of thoracotomy with mobilization of a portion of the fundus of the stomach through an opening in the diaphragm and suturing it to the diverticulum, and the second stage, done four weeks later, to complete the anastomosis. More recently two reports of its successful application have appeared in the American medical literature.[31, 35]

Extirpation of the diverticulum was first performed by Enderlen, in 1910, but the patient succumbed; Stierlein had a similar experience in 1916. Clair-

mont, in 1924, is believed to have performed the first successful extirpation using an abdominal transdiaphragmatic approach. The year before, however, Sauerbruch,[39] in describing a new method of exposing the posterior mediastinum to reach the esophagus, which he termed transpulmonary exposure of the esophagus, reported a case of perforation of an esophageal diverticulum into the lower lobe of the right lung successfully managed by staged operations. Several years later Sauerbruch[40] reported three successful cases, using an anterior mediastinotomy approach in the first, posterior mediastinotomy in the second, and a transpleural approach in the third. This latter approach was used successfully by Meyer to extirpate an epiphrenic diverticulum, according to Torek in 1927. The transpleural approach for extirpation of the sac was also utilized successfully by Fick in 1929, and by Barrett in 1933, and the posterior mediastinotomy approach by Vos in the case reported by Quartero in 1931. More recently a number of surgeons[1, 5, 8, 11, 16, 20, 26, 43, 50] have reported favorably on their experience with this method, the procedure consisting essentially in a transpleural approach through the right or left side, depending on the direction of the pouch, careful mobilization of the sac to expose its neck, complete excision, and repair of the opening in the esophagus by a two- or three-layer anastomosis with closure of the overlying mediastinal pleura.

In an attempt to avoid the development of mediastinitis and empyema, which were the complications feared by the early efforts to extirpate the sac, Lahey,[24] following the suggestion of others[18, 23, 27, 42] for pharyngeal diverticula, advocated and employed diverticulopexy. In this procedure the sac is carefully freed from its surrounding attachments and mobilized upward, and the fundus of the sac is fixed with interrupted sutures to the pleura in the paravertebral gutter so as to afford dependent drainage.

Another form of surgical treatment designed to avoid infection from opening the sac consists in inversion of the diverticulum. This procedure was first employed for pharyngeal diverticula by Girard in 1896. It was also advocated for thoracic esophageal diverticula by Torek, in 1927, who considered it the safest method. It was done by Turner[47, 48] through a posterior extrapleural approach, but the pleura was accidentally injured and the patient subsequently died of pneumonia, empyema, and a bronchopleural fistula.

From an analysis of these various methods for the surgical treatment of epiphrenic diverticula, it would seem that complete extirpation with proper repair of the opening in the esophagus is the method of choice, since it usually achieves the ideal objective, that is, removal of the lesion and restoration of normal anatomy and function. This objective, however, cannot always be achieved by this method, owing to certain pathologic changes consequent to long-standing ulceration, esophagitis, and stricture formation which may extend beyond the neck of the diverticulum to involve the segment of the esophagus just distal to the pouch. Such a condition was encountered in the case to be described herein. Under these circumstances it would appear that resection of the lower portion of the esophagus including the diverticulum and cardia of the stomach followed by esophagogastrostomy provides a practical method for dealing with this condition. Experience with this technique in the treatment

of other lesions of the esophagus has demonstrated its safety and efficacy. Since no reports of the clinical use of this form of surgical treatment for epiphrenic diverticula have appeared in the literature, it seems desirable to direct attention to its employment under these circumstances by presentation of the following case.

REPORT OF CASE

J. B. T., a white man, aged 59 years, was first seen on Sept. 13, 1949, with a history of difficulty in swallowing of nine years' duration. Ingested liquids and solid foods would "hang up" beneath the lower end of the sternum, and occasionally he would vomit an hour or two after meals. The regurgitated food was foul smelling and extremely distasteful. He was frequently awakened at night by a "scratching" sensation in his throat which

Fig. 1.—Oblique roentgenogram of the esophagus following ingestion of barium demonstrating the diverticulum with dilatation and coiling of the esophagus above and narrowing below.

provoked bouts of coughing. Soon after the onset of these symptoms he consulted a physician who, after barium study of the esophagus, made a diagnosis of diverticulum of the lower portion of the esophagus. This condition was treated conservatively with moderate relief of symptoms; however, during the past year the symptoms had increased in severity and serial roentgenograms showed the diverticulum to be enlarging.

Examination revealed a well-nourished and well-developed white man with no gross abnormalities. An esophagogram disclosed a large diverticulum of the distal end of the esophagus arising from the left lateral wall just above the cardiac end of the stomach. Above the diverticulum the esophagus was quite dilated and somewhat tortuous, whereas distal to it a definite narrowing could be demonstrated (Fig. 1). A diagnosis was made of epiphrenic diverticulum of the esophagus with stricture formation, and the patient was admitted to the hospital for surgical therapy.

On Sept. 15, 1949, under endotracheal nitrous oxide and ether anesthesia, with the patient in the right lateral decubitus position, the eighth rib was resected subperiosteally,

and through its bed the left thoracic cavity was entered. Preliminary exploration confirmed the roentgenographic observation of a diverticulum protruding from the left posterior lateral wall of the esophagus just anterior to the aorta and immediately above the cardia. The mediastinal pleura was incised, and the lower third of the esophagus, including the diverticulum, was mobilized. The diverticulum was found to measure approximately 5 cm. in length and 3 cm. in diameter. The esophagus distal to the opening of the diverticulum and extending to the cardia appeared markedly constricted, with a firm thickened wall moderately adherent to the surrounding tissue. In the presence of this rather pronounced degree of chronic esophagitis and stricture formation, it was believed that diverticulectomy alone might not provide adequate relief of the obstructive symptoms. For this reason it was considered preferable to resect the segment of esophagus involved in the chronic in-

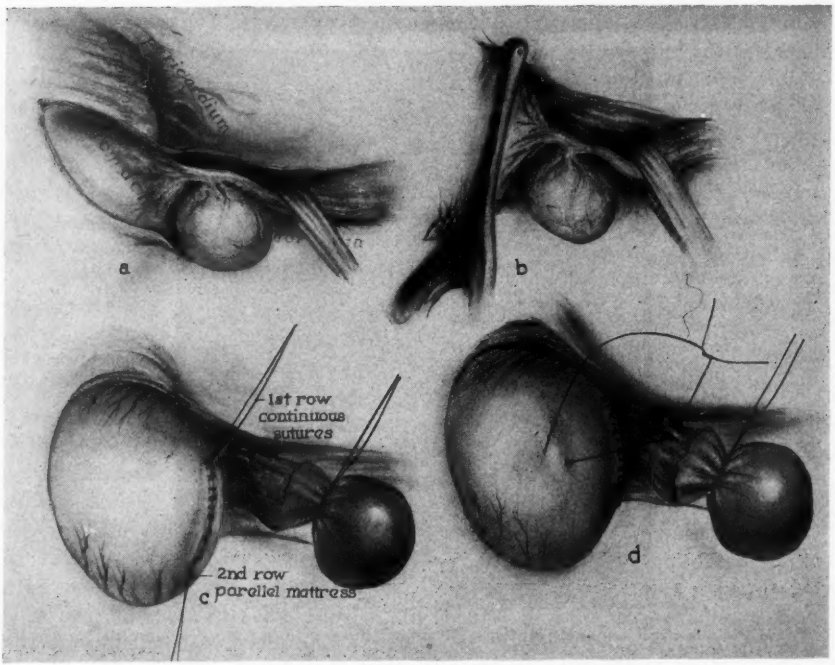

Fig. 2.—Drawing made from authors' case showing (*a*) mobilization of lower esophagus, diverticulum, cardia, and fundus of stomach after opening diaphragm, (*b*) division of stomach just below cardia, (*c*) closure of the gastric stump, and (*d*) beginning esophagogastric anastomosis.

flammatory process along with the diverticulum from just above the opening of the pouch to the cardia and restore continuity by esophagogastric anastomosis. The diaphragm was divided from the esophageal hiatus anterolaterally for a distance of 8 cm. Through this incision the fundus and cardiac end of the stomach were mobilized by division of the vasa brevia and left gastric vessels. After approximately the upper fourth of the stomach had been mobilized, it could be brought up into the chest without difficulty. The stomach was divided at the cardia between Payr's clamps, and the gastric stump closed with a running suture of 000 chromic catgut followed by a row of interrupted Lembert sutures of cotton (Fig. 2). The fundus of the stomach was pulled up into the mediastinum, and esophagogastrostomy was performed utilizing a two-layer anastomosis with cotton (Fig. 3). The stoma was constructed in the share of an L to increase its diameter.[6] The fundus of the stomach was

anchored to the mediastinal pleura and the diaphragm closed about the stomach. A No. 32 Pezzar catheter was placed in the ninth interspace in the posterior axillary line for underwater drainage, and the thoracic wound was closed in layers with cotton sutures. During the procedure, the patient received 1500 c.c. of blood, and left the operating room in good condition.

Examination of the gross specimen revealed an esophageal diverticulum of the pulsion type. Approximately 0.5 cm. distal to the mouth of the diverticulum the esophagus was constricted to an internal diameter of 0.5 cm. with a firm, thickened, and somewhat scarred wall. Histologic examination revealed thickened stratified squamous epithelium lining the

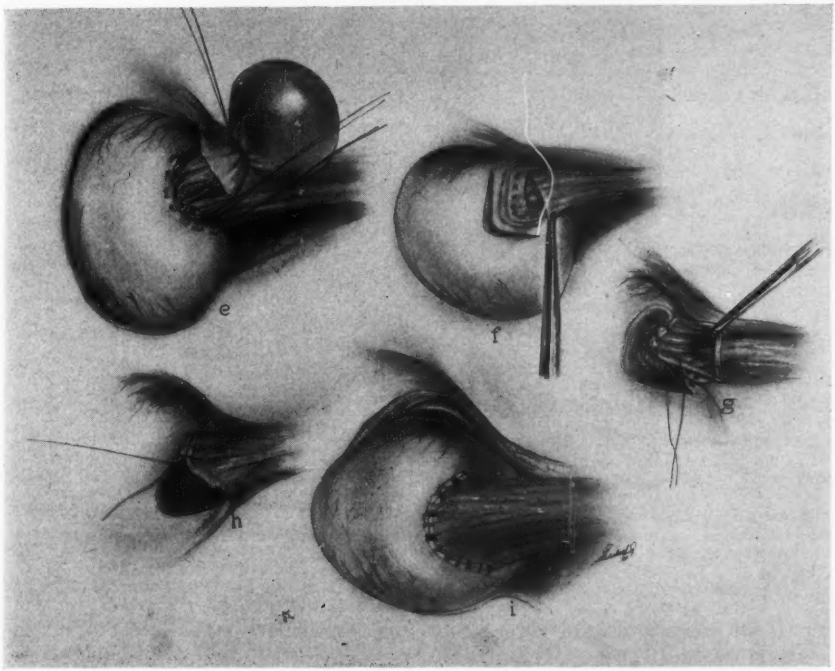

Fig. 3.—Drawing showing steps in technique of esophagogastrostomy performed on authors' case continued from Fig. 2. (*e*) The first posterior row of sutures is placed between the posterior wall of the esophagus and the anterior wall of the fundus of the stomach with an extension upward for about 2 cm. along the left lateral margin of the esophagus. (*f*) The esophagus and stomach are opened by incisions just distal to this first row of sutures, producing L-shaped openings to increase the size of the stoma. (*g*) Placement of inner row of posterior interrupted sutures of quilting cotton, and (*h*) first layer of anterior sutures to provide careful approximation of mucosal edges, care being taken to place sutures so that knots are within the lumen as suggested by Churchill and Sweet. (*i*) Completion of anastomosis by outer layer of anterior sutures.

diverticulum, but no evidence of anaplasia. The remainder of the mucosa for the most part was artifactually absent; however, at one end there was glandular mucosa representing the cardia of the stomach. The smooth muscle layers were well developed in the wall of the esophagus, but were entirely absent in the wall of the diverticulum. Mild focal lymphocytic infiltration was seen throughout the layers of the esophagus. The pathologic diagnosis was pulsion type diverticulum of the esophagus.

The postoperative course was uneventful. The drainage catheter was removed on the third postoperative day, and small oral feedings were begun the fifth postoperative day. The diet was gradually increased, and the patient was discharged ten days after operation.

Following his return home he was able to eat without difficulty, and when examined approximately one and one-half years after resection of the diverticulum the patient was asymptomatic, and barium study of the esophagus at that time revealed an adequate and well-functioning esophagogastric stoma (Fig. 4). When last observed on Sept. 11, 1951, approximately two years after operation, he was still well and asymptomatic.

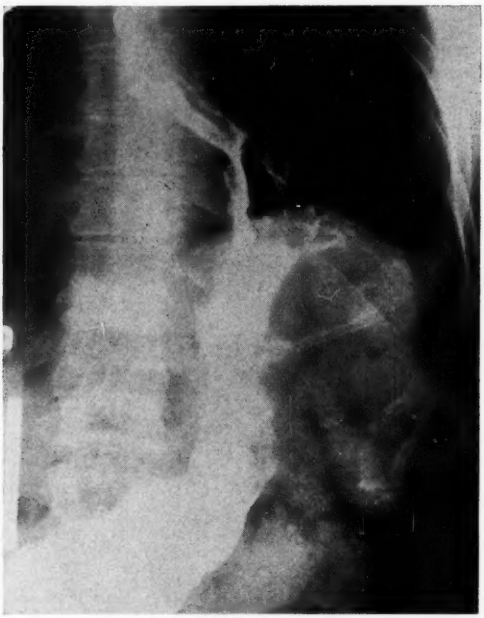

Fig. 4.—Barium study one year after operation showing a normally functioning esophagogastric stoma.

SUMMARY

1. Diverticula of the lower esophagus, particularly those located in the terminal portion and termed supradiaphragmatic or epiphrenic diverticula, are infrequently encountered. Although the pathogenesis of these diverticula has not been clearly established, they are generally considered to be of the pulsion type resulting from herniation of the mucous membrane through the muscular wall, probably in an area of congenital weakness. Symptoms are produced by the occurrence of certain complications, such as partial obstruction resulting from distortion and angulation of the esophagus or inflammation and ulceration with consequent stricture formation as in the case described, or from perforation, hemorrhage, or malignancy.

2. Surgical treatment is the method of choice and should be considered in all esophageal diverticula-producing symptoms. From a review of the literature the procedures that have been advocated and employed in the surgical treatment of epiphrenic diverticula may be classified into the following categories: (1) anastomosis between the sac and the stomach, (2) extirpation of the sac, (3) diverticulopexy, and (4) inversion of the pouch.

3. Complete extirpation of the diverticulum with proper repair of the opening in the esophagus is considered the method of choice. In the case reported this procedure was not considered satisfactory, owing to the presence of marked stricture formation of the esophagus distal to the diverticulum. For this reason, the procedure employed consisted in resection of the lower esophagus including the diverticulum and cardia of the stomach followed by esophagogastrostomy. Although no report has been found of the use of this procedure for this purpose, in light of this experience it is recommended as a practical method for dealing with this condition.

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MISINTERPRETATION OF FINDINGS IN CATHETERIZATION OF ANOMALOUS PULMONARY VEINS ADJACENT TO A MEDIASTINAL DERMOID CYST

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THE diagnosis of a mediastinal dermoid cyst is not always difficult when there is evidence of a progressively enlarging tumor mass in the anterior mediastinum in an individual in the third decade.[1, 2] The recognition of anomalous pulmonary venous drainage, however, depends upon special procedures, since there are no reliable physical signs by external examination of the chest.[3] These procedures include direct inspection of the hilar vessels at either the operating table[4] or the post-mortem table,[5] and demonstration by angiocardiography or by the fortuitous intubation of one or more such veins during venous catheterization of the heart.[3, 6, 8-11] Because the latter approach to a diagnostic problem provided information which was misinterpreted prior to exploratory thoracotomy, the following case is presented:

C. M. J., a 23-year-old white Naval seaman, was referred for diagnostic studies for possible congenital cardiovascular disease. A routine chest roentgenogram one month previously showed an enlarged cardiac shadow (Fig. 1), which was not apparent on a film taken five years earlier (Fig. 2). The possibility of a pericardial cyst was considered also. The patient had been asymptomatic except for occasional palpitations on exertion. He was 6 feet, 3 inches tall, weighed 195 lbs., and appeared to be in excellent health. There was no evidence of cyanosis, clubbing, venous distention, or edema. A slight precordial bulge was present, but there was no marked intercostal pulsation and no thrill. On percussion the area of cardiac dullness extended from the left midclavicular line to a point 4 cm. lateral to the right sternal margin in the third and fourth interspaces. The second pulmonic sound was accentuated, and there was a grade 2 systolic murmur along the left sternal border. There was no diastolic murmur. There was a sinus bradycardia with a rate of 58 per minute; the blood pressure was 135/80 mm. Hg. The remainder of the physical examination was normal.

Chest fluoroscopy, cinefluorography, and radiography revealed clear lung fields with apparently normal bronchovascular markings (Fig. 1). The transverse diameter of the heart was widened, however, due to a pulsating shadow which extended to the right and anteriorly. It was continuous with the cardiac shadow and was observed to diminish in size with the Valsalva maneuver (Fig. 3). In the right anterior oblique view, it extended anteriorly toward the rib cage, thereby simulating an enlarged right heart.

An electrocardiogram showed an intermediate position, or normal axis deviation, and sinus bradycardia of 55 per minute. There was no evidence of a conduction defect or ventricular hypertrophy, and the tracings were within normal limits. The hemoglobin concentration was 15.1 Gm. per cent (Van Slyke determination). The urine was normal. An exercise tolerance test using a 10-minute step-test technique[12] was entirely normal; the physical

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This investigation was supported in part by Research Grant H-908(R) from the National Heart Institute of the National Institutes of Health, Public Health Service.

Received for publication Oct. 29, 1951.

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TABLE I

LOCATION OF CATHETER TIP	O_2 SATURATION (%)	PRESSURES (MM. HG)
Superior vena cava	73	--
Inferior vena cava	82	--
Right auricle	77	7-10*
Region of the mass	93	22-25*
Right ventricle	--	30/2-5
Pulmonary artery	79	26/15
Femoral artery	96	--

*Variation in mean pressure with respiration.

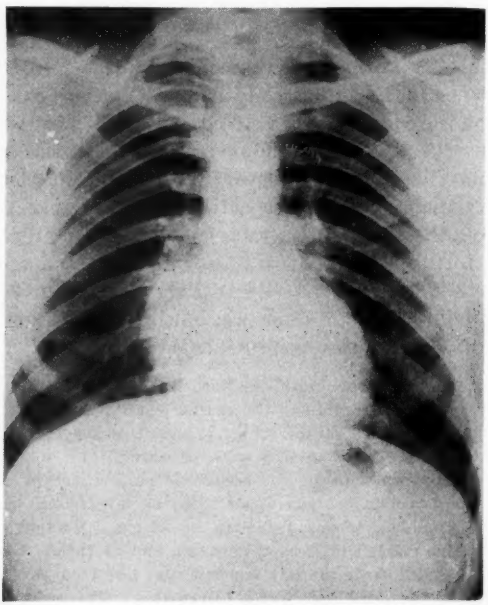

Fig. 1.—Chest roentgenogram of C. M. J. taken in December, 1950. Note the unusual contour of the right border of the cardiac shadow.

fitness index was 22.8. There were no abnormalities in the chest-lead electrocardiogram during the exercise. Venous catheterization of the right heart yielded the information noted in Table I.

Since the oxygen consumption was 320 ml. per minute, and the AV oxygen difference was 37 ml. per liter, the cardiac output at rest was a high normal value of 8.65 liters per minute. The stroke volume was 129 milliliters. Total pulmonary resistance was estimated to be about 157 dynes sec. cm.$^{-5}$, whereas that of the periphery was about 906 dynes sec. cm.$^{-5}$. The tip of the catheter was inserted into an anomalous vessel entering the superior vena cava (Fig. 4) where highly oxygenated blood was withdrawn. The mean pressure at this point (22 to 25 mm. Hg) was higher than that observed for either the right auricle (7 to 10 mm. Hg) or the left pulmonary artery (14 to 17 mm. Hg), and rose to 60 mm. with performance of the Valsalva test. A somewhat damped arterial pressure pattern was recorded during the Müller test, with a mean pressure of around 30 mm. Hg. Accordingly this vessel was considered to be a communication with one of the systemic arteries. With moderate leg exercises, the pressure in the left pulmonary artery rose to 36/16 mm. Hg.

The observations reported above confirmed the findings of the referring physician that the patient had an abnormal configuration of the heart by roentgenogram examination. The possibility of a pericardial cyst seemed unlikely because of the rather striking reduction in size with the Valsalva maneuver, which was attributed to the effects of greater intrathoracic pressure on venous return to the heart. Insertion of the catheter tip into the region of the

Fig. 2.—Chest roentgenogram taken in July, 1945. Despite the slight convexity of the right auricular shadow, it was considered to be normal.

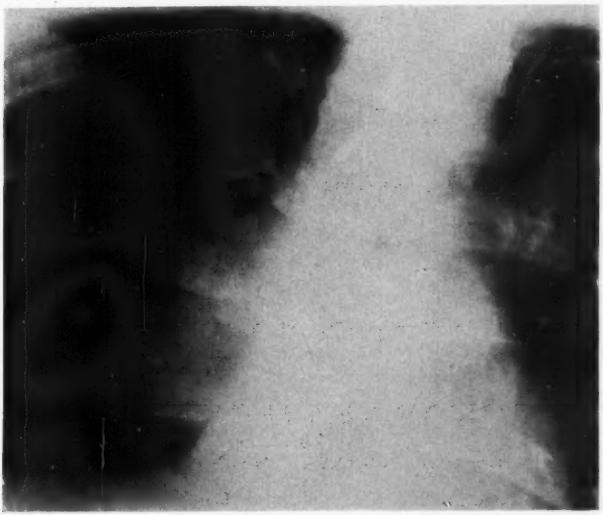

Fig. 3.—Spot film of the right side of the heart taken during cardiac catheterization to demonstrate the reduction in the size of the mass during the Valsalva maneuver. (Compare with Fig. 4.)

mass during venous catheterization of the heart provided three observations of interest: there was an anomalous vessel entering the right auricle, it contained blood that was highly oxygenated, and the blood pressure was appreciably higher than that of the right auricle, increasing to 60 mm. of mercury with the Valsalva maneuver. The pressure pulse which was recorded graphically resembled an arterial pattern. Although the diagnosis was uncertain, the combined findings of physical and roentgenogram examinations, together with the cardiac catheterization indicated an arterioauricular aneurysm or an angiomatous tumor with an auricular communication. Because of the appearance and rapid enlargement of this mass within a five-year period (see Figs. 1 and 2), surgical exploration was considered mandatory.

A right exploratory thoracotomy was carried out on March 22, 1951. Upon entering the chest, a large dermoid cyst was identified. It was attached to the pericardium over the right auricle and the ascending aorta by a broad pedicle. The mass (Fig. 5) was removed without untoward incident (Fig. 6). In order to explain the cardiac catheterization findings, a dissection of the pulmonary hilus was undertaken. The pulmonary venous drainage of the

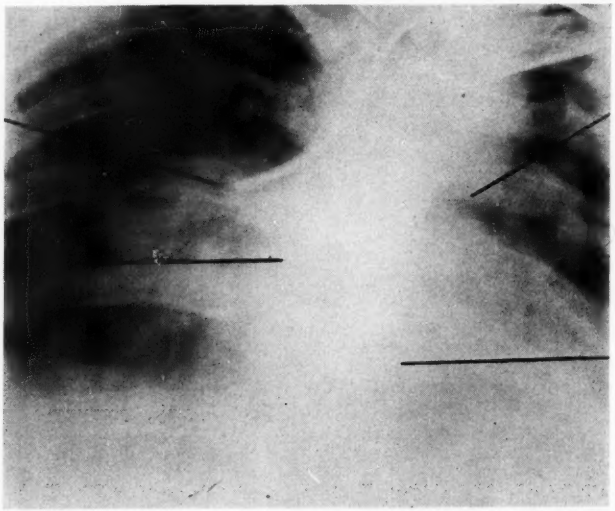

Fig. 4.—Spot film of the heart taken during cardiac catheterization. Note the tip of the catheter in the region of the mass. Oxygen saturations and blood pressures in mm. Hg as indicated.

right lung consisted of three veins. A single large vein draining the right upper lobe emptied directly into the superior vena cava approximately 8 cm. above the pericardial reflexion. A smaller vein from the right middle lobe emptied anteriorly and directly into the right auricle. The inferior pulmonary vein was noted to be in its normal position. No further attempt to trace the course of this latter vein was made.

The patient's postoperative convalescence was uneventful. He was discharged on the twelfth postoperative day completely ambulatory and without complaint. He has since returned to full-time work.

DISCUSSION

Had not cardiac catheterization been performed on this patient, the presumptive diagnosis would have been dermoid or pericardial cyst. The history of palpitations and the fluoroscopic evidence of a pulsating mass, as observed in this patient, have been described in previous cases of dermoid cysts. Venous

pulsations and variations in density with either the Valsalva or Müller experiments have not been striking by roentgenoscopic examination in patients with anomalous pulmonary veins.[7] The preoperative venous catheterization of the heart in this patient, however, provided information which was misinterpreted.

Fig. 5.—The mass measured 7 × 11 cm. The outer surface was golden yellow and somewhat nodular. On section a cheesy material was present intermingled with hair. Microscopic examination revealed only ectodermal elements. The final diagnosis was dermoid cyst.

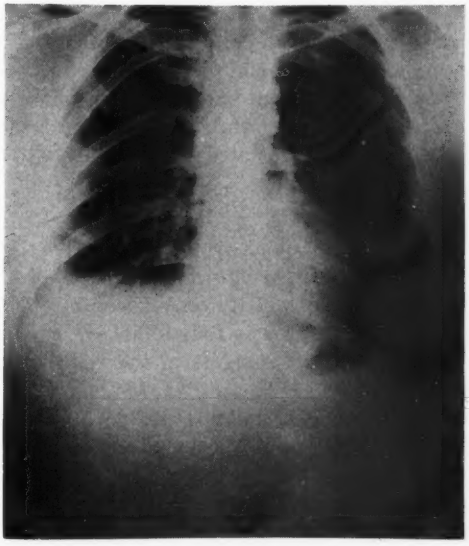

Fig. 6.—Chest roentgenogram taken ten days after removal of the dermoid cyst. The vascular markings below the right hilus are more conspicuous since the overlying tumor has been removed.

This was due to the fortuitous insertion of the catheter tip into one of the anomalous pulmonary veins, the withdrawal of highly oxygenated blood, and observing an appreciably higher pressure than in the adjacent right auricle. Possibly a lateral radiograph would have been helpful in localizing the position of the catheter in relation to the mass, but even this could have been misleading because of the fact that the mass was attached to a pedicle.

Although nearly 300 cases of mediastinal dermoid cyst have been reported,[1, 2] apparently none have been associated with anomalous pulmonary venous drainage. In this patient, the venous anomaly was not recognized until the hilar structures had been dissected carefully at the operating table. Anatomists have found the incidence of anomalous pulmonary venous drainage to be about 0.7 per cent.[13] This patient represents the fourth example of this anomaly that the authors have encountered within the past year. Hence these findings suggest that it is more common than usually stated. Unless pathologists are alerted to the possibility, it is likely that the exact pathology will be overlooked in the type of autopsy routinely performed.

When less than 50 per cent of the pulmonary venous return enters the right auricle the patient has no symptoms,[3, 6] there is little likelihood of decompensation, and the life span need not be shortened.[5] With total venous drainage of the lungs into the right heart, there must be either a septal defect or patency of the ductus arteriosus in order to supply the major circulation with blood that has been oxygenated. In these cases not only is the right ventricle hypertrophied, but the left heart is smaller than usual.[5] Closure of the ductus arteriosus diminishes the blood flow to the greater circulation, leads to dilatation and failure of the right heart, and accounts for the death of the patient in early infancy.[5] With only partial anomalous venous drainage, either collapse or removal of part or all of the lung with normal venous drainage is dangerous.[3, 4, 6] Even atelectasis of the normal lung may be hazardous to the patient.[6] It has been suggested that some of the immediate accidents following pneumothorax may be attributed to such a mechanism.[3] Under any of these circumstances the reduction in the amount of oxygenated blood available to the greater circulation becomes critical.

SUMMARY

1. A report is made of a patient who, primarily on the basis of cardiac catheterization studies, was regarded clinically as having an arterio-auricular aneurysm or an angiomatous tumor with an auricular communication. However, upon surgical exploration he was found to have a dermoid cyst in the mediastinum and anomalous veins going from the right lung to the superior vena cava and right auricle.

2. The danger of any surgical therapy of the lung with normal venous drainage, in the presence of anomalous venous drainage of the opposite lung, is emphasized again.

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CASE REPORT—AN UNUSUAL MEDIASTINAL CYST

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THIS case of a mediastinal cyst is presented because of the difficulties experienced in diagnosis together with an unusual response to certain therapeutic agents.

The patient, Mr. S., a 54-year-old white man, was apparently in good health until February, 1948, when he suddenly lost his voice, being unable to speak above a whisper. Approximately one week later he developed a mildly productive cough with mucoid sputum, once blood-streaked. Approximately two weeks later he noted a numbness and stiffness of the fourth and fifth fingers of his left hand, but this symptom gradually disappeared within a few days. There were no associated weight loss, dyspnea, fever, or difficulty in swallowing. The past history was noncontributory, except for treatment for syphilis in 1931 and again in 1941.

When first seen in March, 1948, the physical examination was negative except for hypertension (200/98) and a complete paralysis of the left true vocal cord. Laboratory studies, including a blood Kahn test, were all within normal limits. An upright posteroanterior and left lateral film of the chest revealed a hazy shadow of uniform density along the right superior mediastinum and extending out to the left for a distance of 2.5 cm. from the lateral border of the spine near the aortic knob. The lateral border of this opacity had a smooth contour. Esophagograms revealed a compression of the esophagus in the region of the fourth and fifth thoracic vertebrae, with a slight displacement of the upper esophagus and trachea toward the right just within the thoracic inlet. Esophagoscopic examination revealed a slight compression of the left wall of the esophagus in the upper third. The esophageal mucosa appeared smooth and normal throughout with no stenosis or intrinsic tumor visualized. Bronchoscopic examination showed the trachea to be compressed on its left lateral surface with a shift toward the right in the same location. The remainder of the bronchial tree was normal. A bronchial washing of the left main bronchus was reported by the pathologist to be negative for tumor cells.

As a result of the above findings, we considered the diagnosis in this case to be a bronchiogenic carcinoma with invasion of the left recurrent laryngeal nerve, and therefore not curable surgically. Since the patient's only incapacity at the time was his weak voice, it was our feeling that he should be treated conservatively for the present, and therefore we withheld Methyl-bis and x-ray therapy until needed. It was decided that an exploratory thoracotomy and biopsy would only cause the patient further discomfort and not add materially to the understanding of his case.

The patient continued to work until January, 1949, when he was admitted to the Baptist Memorial Hospital in Memphis because of severe dyspnea, cyanosis, fever, and peripheral circulatory collapse. Physical examination revealed a fever of 102° F., a pulse rate of 120 per minute, respiratory rate of 26 per minute, a complete paralysis of the left vocal cord, and moist râles throughout both lungs. X-ray examination of the chest (Fig. 1) showed no apparent increase in the mediastinal mass. There was, however, a marked pneumonitis involving the bases of both lung fields. Treatment consisted of nasal oxygen, intravenous fluids, sedation, and penicillin intramuscularly. The respiratory difficulty gradually subsided, and within approximately ten days he was afebrile and able to do without oxygen. On Jan. 26, 1949, Methyl-bis therapy was instituted, 10 mg. being given daily

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Received for publication Nov. 1, 1951.

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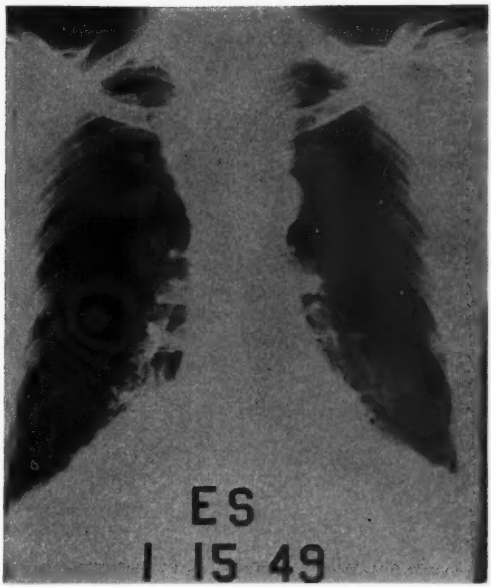

Fig. 1.—Patient E. S., Jan. 15, 1949. There is a hazy shadow of uniform density along the right superior mediastinum which extends out to the left for a distance of 2.5 cm. from the lateral border of the spine near the aortic knob. The lateral border of this opacity has a smooth contour. There is also some pneumonitis involving the bases of both lung fields.

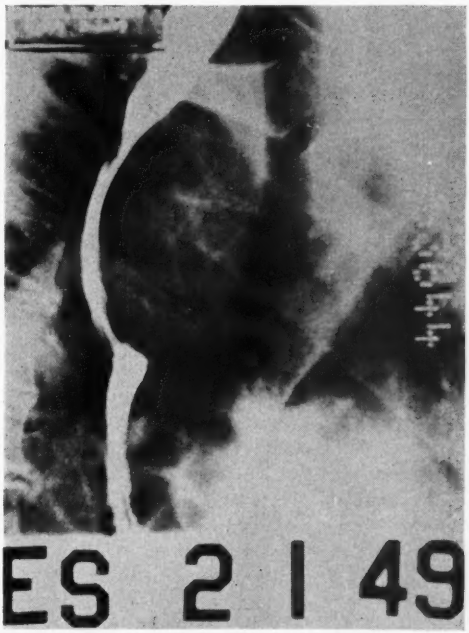

Fig. 2.—Esophagogram Feb. 1, 1949. This reveals a smooth compression of the esophagus in the region of the fourth and fifth thoracic vertebrae with slight displacement toward the right of the upper esophagus and trachea just within the thoracic inlet. The tumor is definitely between the trachea and the esophagus. The smooth contour of the mass suggested a benign neoplasm, but we discounted this possibility because of the presence of the paralyzed vocal cord and were thus led astray for some time.

intravenously for four consecutive days. Following this the patient showed considerable symptomatic improvement, exhibited by more comfortable breathing, and a lessening of his severe cough. A roentgenogram of the chest with barium in the esophagus on Feb. 1, 1949 (Fig. 2), showed the esophagus to be displaced backward, and the trachea displaced to the right and forward. A long fusiform mass with a smooth contour separated the trachea and esophagus. No irregularity was evident in the contour of the esophagus or trachea. A kidney-ureter-bladder film revealed no enlargement of the liver, spleen, or kidneys. Films of the lumbar spine, pelvis, and hips were reported as negative.

A repeat bronchoscopic examination was done on Feb. 7, 1949, which revealed the trachea to be displaced anteriorly. Considerable mucopurulent sputum was observed coming from the orifices of all lobes. A bronchoscopic sputum specimen was taken, and this was

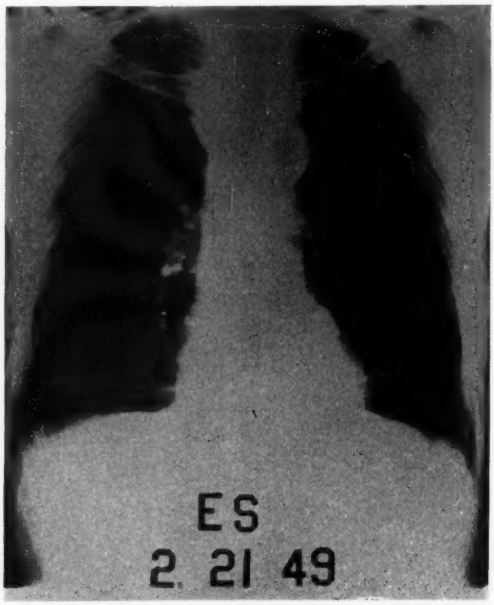

Fig. 3.—Feb. 21, 1949. After a course of Methyl-bis intravenously, 10 mg. once a day for four days, and intensive x-ray therapy to the superior mediastinum, there has been a marked decrease in the size of the tumor along the right superior mediastinum. This again confused us and made us believe that we were dealing with a bronchiogenic carcinoma or lymphoma as we have seen a similar response in this region in other cases.

reported as being negative for tumor cells. A repeat posteroanterior film of the chest taken on Feb. 8, 1949, revealed a *sharp diminution in the size of the mediastinal mass* and a clearing of the pneumonitis in both bases. The patient was placed on active bronchial drainage routine with ammonium chloride and discharged home on Feb. 9, 1949, with instructions to return within ten days for an exploratory thoracotomy. This procedure was now deemed advisable since there was a good possibility the mass was benign, because of the smooth borders of the tumor, and because of the lack of invasion of contiguous structures (except for the left recurrent laryngeal nerve) for the period of time the patient had been under observation. Upon his return to the hospital on Feb. 21, 1949, a posteroanterior roentgenogram of the chest (Fig. 3) showed the mediastinal mass to have almost completely disappeared. Because of this dramatic response to Methyl-bis therapy it was now thought that this

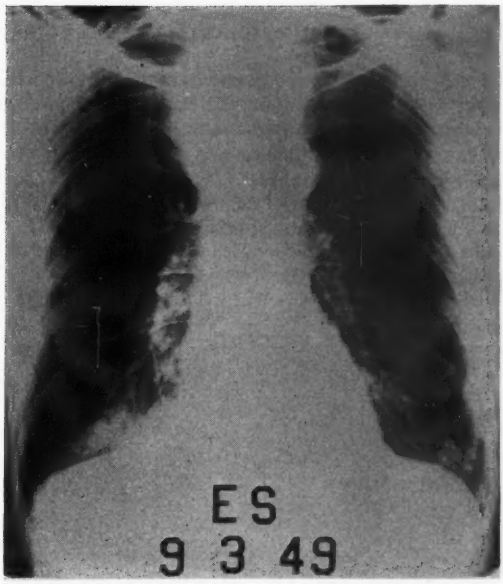

Fig. 4.—Sept. 3, 1949. The patient had fever and respiratory difficulty at this time and roentgenograms revealed an increase in the size of the neoplasm in the right upper chest, and again a pneumonitis in the right cardiophrenic angle. After the pneumonitis was controlled, an exploratory thoracotomy was decided upon, but was refused by the patient for some time.

On Oct. 1, 1949, operation was performed and a large tumor resected. This tumor occupied the triangle between the superior vena cava anteriorly, the azygos vein inferiorly, and the esophagus posteriorly. The cyst was removed in toto.

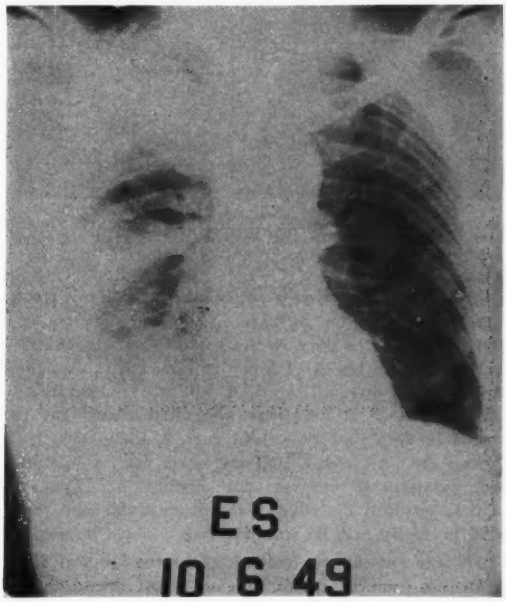

Fig. 5.—Oct. 6, 1949. Roentgenogram made a few days after operation reveals some pleural reaction over the entire right lung field but the tumor has been removed completely from the right superior mediastinum. The patient's convalescence since this time has been most gratifying and he is now back at full-time work.

was a case of lymphoma, either of the lymphosarcoma or Hodgkin's class, and not curable by surgery. Therefore the patient was again discharged, and instructed to return as an outpatient for x-ray therapy to the upper mediastinum.

The patient was admitted again to the hospital on April 15, 1949, with an acute respiratory infection. Temperature was 101° F., pulse rate was 104 per minute, and respiratory rate was 36 per minute. He exhibited signs of partial tracheal compression with associated bilateral decreased breath sounds. Posteroanterior roentgenogram of the chest showed the mediastinal mass to have resumed its former size and contour. The patient was again treated symptomatically and given 40 mg. of Methyl-bis in divided doses. One week from the date of admission he was feeling well, and was discharged home, ambulatory.

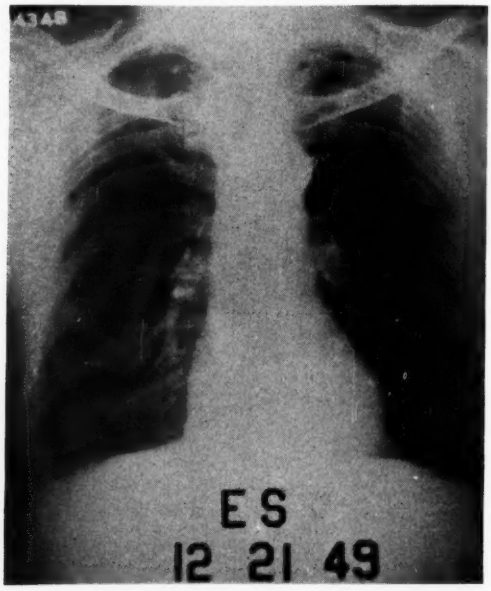

Fig. 6.—Follow-up roentgenogram, Dec. 21, 1949, shows marked clearing of the pleuritis on the right. The patient is asymptomatic except for hoarseness, and the paralyzed left vocal cord is still unchanged.

During May, 1949, the patient was given a second series of x-ray treatments to the upper mediastinum. However, he continued to have a severe productive cough with tenacious yellowish sputum and dyspnea, especially in the supine position. Since he received no immediate benefit from the second series of x-ray treatments, it was decided to try to keep the patient comfortable on Methyl-bis injections (10 mg.) intravenously every two weeks in the office, and to give blood transfusions when indicated. The patient continued to breathe comfortably and to work part time until Sept. 2, 1949, when he was admitted again to the hospital with extreme respiratory difficulty, fever, and a bilateral basal pneumonitis (Fig. 4). Treatment consisted of oxygen, aminophylline, and 5 per cent dextrose intravenously twice each day, postural drainage, and intramuscular and aerosol penicillin. The pneumonitis responded promptly and he was discharged home on Sept. 11, 1949.

On Sept. 28, 1949, the patient was readmitted to the hospital for a right exploratory thoracotomy, as our doubts grew as to his diagnosis. At operation a large cystic tumor, approximately 10 cm. in its longest diameter, occupied the triangle between the superior vena cava anteriorly, the azygos vein inferiorly, and the esophagus posteriorly. The cyst

was removed in toto. Its contents were a thin crystal-clear watery secretion. The day following operation an emergency tracheotomy was necessary because of the paralyzed vocal cord, and edema of the vocal cords after endotracheal anesthesia. Otherwise, the postoperative course was uneventful. The tracheotomy tube was removed in a few days and the opening allowed to close by granulation. The patient was discharged home on Oct. 14, 1949, afebrile and asymptomatic, except for hoarseness (Fig. 5).

The cyst was reported by the pathologist to be a mesothelial-lined cyst compatible with a cavernous lymphangioma.

The patient was last seen on Aug. 7, 1950, at which time he was working full time with no symptoms referable to his chest.

SUMMARY

A benign mediastinal cyst resembled clinically a bronchiogenic carcinoma with mediastinal invasion. The cyst decreased remarkably in size with x-ray therapy and Methyl-bis injections especially the latter. This decrease in size could be attributed to the vomiting and dehydration caused by the nitrogen mustard, or else due to shrinkage of the lymphatics that fill the cyst due to radiation therapy.

Having learned a lesson from his case, we now believe that exploratory thoracotomy is indicated for the patient with supposed bronchiogenic carcinoma with a paralyzed vocal cord, unless definite evidence of tumor exists elsewhere.

AN ACCESSORY LUNG

Report of a Case

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ANOMALIES of the tracheobronchial tree are numerous. However, there have been few reports in the literature of a true accessory lung. The so-called accessory lung can be classified into two main types: the lung generally stemming from the lower lobe, and the lung which derives directly from the trachea. It is preferable to call the lung stemming from the lower lobe an accessory lobe. Gruenfeld and Gray,[6] reviewing the literature, cited Herxheimer[7] and Müller,[12] each reporting a case of true accessory lung in which the main bronchus came directly off the trachea. The first case was reported by Herxheimer in 1901, and Müller reported his case in 1918. In 1940, Hulse[8] reported a case of a true tracheal, accessory lung which was diagnosed by bronchogram. Our case fits into the Freedlander-Gebauer[5] classification as a tracheal accessory lung. It differs from the case reported by Hulse, as it was not only small masses of lung tissue but a whole lung. It was not situated in the mediastinum, but in the right thorax. Careful review of the English literature shows our case to be the first of a true accessory lung removed by surgery. Von Haberer[15] removed a cyst from the right interlobar fissure. Postoperative roentgenographic studies showed a bronchus leading to the region where the cyst was removed. He assumed that he had removed an accessory apical lung. In reviewing the embryology of the tracheobronchial tree, one finds many reasons for lung abnormalities. As early as in the fourth week of embryonic life, the posterior end of the outpouching arising from the caudal part of the respiratory bud assumes a bifid form. The lung buds are asymmetrical almost from the very beginning, the right being larger than the left and more caudal. The bronchial tree is formed by repeated division of the lung bud in its branches. This multiple division of the bronchial tree from the original bud gives rise to many abnormalities. Lung anomalies occur when the tracheal buds push into the mesenchyma and become partially isolated. Santy, Bérard, and Galy[13] feel that all cysts having origin in the region of the pulmonary pedicle arise from an accessory bronchial bud, or from a supranumerary lobe. In our case a complete lung with three lobes arose from a main bronchus which came directly off the trachea.

Case History

L. M. B., a white female infant, two months old, was seen because of a persistent cough. Chest roentgenogram revealed a homogeneous density occupying the upper two-thirds of the right hemithorax and suggesting atelectasis of the right upper lobe. Also anomalies of

Received for publication Nov. 5, 1951.
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the lower cervical and upper thoracic vertebrae were noted with incomplete fusion and deformity. The first symptoms appeared two days after birth. There was a history of multiple congenital anomalies on the father's side. The mother had no communicable or other diseases while pregnant, except for an upper respiratory infection with fever during the fourth month. The patient also presented a hip deformity, with the left leg being shorter than the right.

A bronchogram was advised and performed. The bronchogram showed a stem bronchus coming directly off the bronchial tree to the right and saccular bronchiectasis in the area of the density occupying the upper two-thirds of the right hemithorax. At this time a

Fig. 1.—Bronchogram on Jan. 23, 1949, shows a bronchus coming directly off the trachea and saccular bronchiectasis of the area occupied by the density. Another bronchus is seen coming off the trachea below the upper bronchus.

Fig. 2.—Chest film taken on May 10, 1950, shows opacification of upper three-fourths of right thorax.

diagnosis of a saccular bronchiectatic right upper lobe was made, and surgery was advised. Due to the patient's condition, conservative treatment was advised until she was older and in better condition for operation.

On May 9, 1950, the patient was readmitted to the hospital because her symptoms were worse, and fever had appeared. She could not take her feedings. Coccidioidin skin

Fig. 3.—Roentgenogram taken on July 13, 1950, just prior to operation reveals essentially no change.

Fig. 4.—Postoperative roentgenogram revealing complete filling of right thorax by the remaining lung.

test and Mantoux test were negative. The urine showed traces of albumin. The patient received aureomycin, improved, and was discharged on June 17, 1950. She was readmitted on July 27, 1950. Her condition became worse, and she was choking easily. Surgery was advised, and a right exploratory thoracotomy was performed on July 28, 1950.

Upon opening the right pleural cavity, a large and hard tumor mass, apparently consisting of lung parenchyma, was seen and felt occupying three-fourths of the entire hemithorax. This was thought to be a cystic lobe; however, careful inspection revealed this lung to be in its own pleural cavity completely separated from the other right-sided lung which

was situated inferiorly and was partially collapsed. This lower lung had a normal appearance, but it had only two lobes. It also had its completely separate, pleural cavity. The upper, accessory, cystic lung came directly off the trachea by a stem bronchus that measured approximately 3 mm. in diameter and 1 cm. in length. Two small veins each measuring 1 mm. were seen leaving the lung and following the bronchus. The arterial supply was furnished by a branch coming directly off the subclavian artery and entering the cystic lung at its apex. This artery measured approximately 2 mm. in diameter. There was no other blood supply to this lung. A right upper complete pneumonectomy was performed by the individual ligation technique. The upper parietal pleural sac was completely excised, and

Fig. 5.

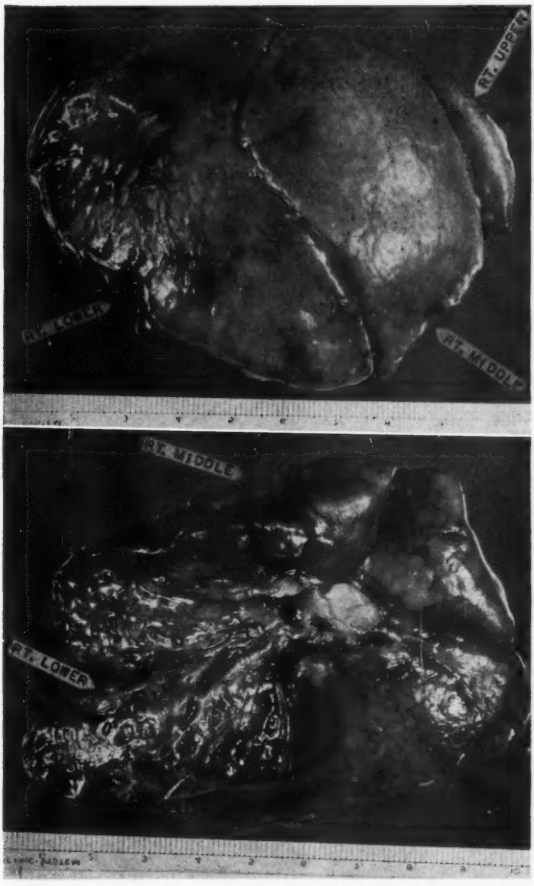

Fig. 6.

Fig. 5.—Tracheal accessory lung removed with three separate lobes, two complete fissure lines.

Fig. 6.—Cut section of one of the lobes shows the nature of the cystic parenchyma.

when the anesthetist applied positive pressure, the lower right lung completely expanded and filled the entire right hemithorax. A catheter was left in place for suction, and the chest was closed in a routine manner.

The patient had an uneventful convalescence, her respiratory complaints disappeared, and she gained weight steadily. Due to some genitourinary symptoms a retrograde pyelogram was done on Sept. 18, 1950 which revealed bilateral hydronephrosis with right hydroureter and a contracture of the vesical neck. The patient was discharged in apparently good health on Oct. 18, 1950, and was followed in the outpatient clinic. She was readmitted on Dec. 17, 1950, and a cystotomy with transverse resection of the vesical neck was performed on Jan. 10, 1951. The patient was discharged on Jan. 19, 1951, and at the present time is symptomatically and clinically well. A chest roentgenogram on Dec. 26, 1950, showed no active pathology, with both hemithoraces and lungs being normal. The patient was last seen on July 20, 1951, while admitted to the hospital for a genitourinary check-up. She is now free from respiratory symptoms.

The pathologic specimen consisted of a lung with three lobes, covered by an intact pleura. The lung weighed 82 Gm. and measured 9 by 7 by 3 centimeters. Two complete fissure lines were present. The specimen had a hilum with three major bronchi and three blood vessels. The parenchyma was filled with cystic spaces, the bronchioles were dilated, and a considerable amount of honeycombing was present.

SUMMARY AND CONCLUSION

A case of a true accessory lung is reported.

A brief review of the embryology of the tracheobronchial tree is given.

Review of the English literature shows this to be the first case treated with success by surgical excision.

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CARCINOMA OF INTRATHORACIC TRACHEA: EXCISION AND REPAIR BY TANTALUM GAUZE-FASCIA LATA GRAFT

REPORT OF A CASE

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THE repair of defects of the intrathoracic trachea has recently been the subject of intensive clinical and laboratory study. Experimentally, defects have been created to simulate conditions in the human being, in whom sections of the trachea are lost after trauma, or excised as part of the treatment of strictures and tumors arising in, or secondarily, invading the trachea.

From the few successes and the many failures of these various studies, two major problems have become apparent: the maintenance of anesthesia and oxygenation during the repair, and the reconstruction of the segment to be excised so that the trachea may resume physiologic function.

ANESTHESIA

One of the most trying problems has been to devise a method of maintaining an adequate airway distal to the area being repaired, for, as the defect is made, there is a loss of the effective intraluminal pressure, as well as an escape of oxygen and anesthesia. To be sure, this situation obtains, to some extent, during the sectioning of a bronchus at pneumonectomy. But bronchial closure in that procedure is rapidly made as the lumen is opened, whereas a large defect in the trachea requires a much longer time for reconstruction.

This difficulty has been circumvented in various ways. The most obvious method is to make use of the already indwelling endotracheal tube which is in place proximal to the area to be excised. As the defect is created, the balloon on the tube is deflated and, by the combined efforts of the anesthesiologist advancing the tube and the surgeon pulling it down through the defect, the tube can be passed across the aperture and manipulated into the desired bronchus. The balloon can them be reinflated. The weakness of this method became apparent in the case to be described, when the distal tip of the tube entered one bronchus alone, thereby making difficult the inflation of the opposite lung when this was needed (usually on the operative side). In attempts to overcome this, if the tip of the tube is suspended just above the carina so as to feed both bronchi, the average balloon will not hold on a short tracheal wall cuff and may fall into the excised area, and the system would no longer be airtight. This difficulty was overcome by Belsey[1] when he added an elongated balloon to the tube to fill the excisional defect while stainless steel wire was wrapped about both balloon and trachea—which is used in the manner of a darning egg, and then covered by fascia lata.

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Presented before The New York Thoracic Surgical Society, Feb. 16, 1951.
Received for publication Nov. 7, 1951.

Carter and Strieder[2] have described what may prove to be the most practical idea. At the time of thoracotomy, rubber tubes of varying diameters and lengths are available and are inserted into the lumen through the defect until a snug fit is achieved. The tube, in place, overlaps the proximal and distal wound edges and serves as a temporary bridge while the repair is carried on about it. At the conclusion of the procedure it is removed by the bronchoscope. Perhaps if plastic or stainless steel tubes were used in this manner, they might be left in place for one to two days and serve the additional function of splinting the repaired area providing a readily available airway and exit for secretions, until sealing of the peritracheal tissues had begun. This additional tube could then be removed endoscopically.

The insertion of an oxygen-containing catheter or another endotracheal tube through the chest wall wound into the defect is of questionable value; it encumbers the surgeon, and the tidal exchange of gases in a lengthy tube extension is inadequate and may produce anoxemia.

PRINCIPLES OF REPAIR

Certain fundamental principles of repair peculiar to the trachea have become apparent, and if any technique or material can be judged to be competent, it must fulfill most of the requirements of these principles.

The means of repair employed should vary with the condition that requires it; no single method covers every type of situation. Thus, small defects, particularly of the membranous portion of the trachea, can be bridged well by fascia lata or pleura sutured tautly over them.[7] This measure is possible as long as there is skeletal supoprt of the surrounding tracheal wall sufficient to maintain a good luminal diameter. However, large defects and excision of the entire circumference of the tracheal wall would not be corrected in this manner because of the "flutter"—or instability—of the graft when special stress is placed on the tracheobronchial tree, as in deep breathing or coughing. Moreover, it does not serve as a bulwark against the constricting effect of subsequent fibrosis or the encroachment of neighboring structures.

If no more than two or three cartilaginous rings have to be excised as, for example, in the treatment of a benign stricture, sleeve resection and end-to-end anastomosis after mobilization of the trachea has been carried out.[4] But should longer lengths of trachea require excision, there is an ever increasing danger of strain on the suture line which may separate and result in disaster. Thus, for large benign tumors or carcinomas which must have a wide margin of normal tissue to insure total excision, these limited means would be inadequate, and under the pressure of securing a closure, the surgeon might be tempted to leave a dangerously narrow cuff about disease.

As the defect enlarges, the problems of its repair increase apace, and due consideration must be paid to certain principles which we have classified as being of *immediate* and *permanent* concern.

For example, it is of *immediate* importance that the material used be relatively easily managed; no undue tension should be placed on the suture line on the trachea for fear of eventual disruption; an air- and watertight

seal must be effected (air prevented from leaking out and fluid from leaking in from the surrounding structures). If this is not entirely achieved at operation it should certainly supervene within a day or two; bacteria should be kept to a minimum; and secretions must be removed adequately during the postoperative period until the tracheal defect has become well sealed off and the patient competent in this regard.

It is of *permanent* concern that re-epithelization of the defect occur and an adequate airway be maintained for the rest of the patient's life. The trachea can be considered as a "drain as well as an air duct, depending upon the ciliated epithelium to empty itself." That method of repair has merit that takes into consideration and utilizes the remarkable ability of ciliated epithelium to regenerate and spread itself across large surfaces if supported from beneath.

However, although epithelization is complete, it provides no bulwark of itself to the threat of delayed luminal narrowing as a result of scarring. This must be anticipated by providing a rigid mural support, tolerated by the body, for an indefinite length of time, for the surrounding tissues about the intrathoracic trachea cannot be relied upon to maintain the luminal diameter. Cartilaginous rings show signs of regeneration many months after the procedure by which time irreversible stenosis may well have eventuated. These rings are frequently of primitive type and may never achieve a mature contour needed to maintain a good lumen.

Various materials satisfy in part these immediate and permanent requirements as set forth above. Stainless steel and plastic tubes are easily inserted, provide a good lumen while they are in place and are tolerated surprisingly well for a fair length of time.[3] However, when re-epithelization occurs it has been found to progress *behind* these tubes so that their intraluminal surfaces are physiologically inert. If, by chance any intercurrent irritation or infection requires the removal of the tube, a debilitating stenosis at the site of repair almost always supervenes. In addition, in those instances in which carcinoma has been present, the tube in place may cover an early local recurrence of disease that might otherwise be easily observed endoscopically.

At present, work with autogenous and homogenous grafts has revealed these to be of questionable value in tumor cases. When the former are used, at least a resection of lung tissue is necessary so that a portion of uninvolved bronchial wall for grafting can be obtained to repair partial defects of the trachea.[2] However, the transplant so derived may be from an infected and/or carcinomatous lung and may possibly serve as a vehicle to return these contaminants to the patient. The homogenous grafts have been used experimentally for circumferential defects.[5] Although fresh and preserved specimens are easily handled, when defects of four or more rings are replaced by this method, late fibrosis and stenosis result and often cause the death of the animal.

For over-all utility and results in repairing large defects, from the evidence derived from human experience, the use of a metallic framework lined by a fascia lata, dermal, or pleural graft seems most promising. Belsey has had experience with the use of stainless steel wire loops, about which fascia

lata has been wound. A primary tumor of the intrathoracic trachea was excised, and the defect managed by this technique. In another instance, a portion of trachea and carina was excised and repaired with apparently good results. Although, functionally, these repairs were adequate, 40 per cent luminal narrowing was estimated to have occurred after a period of several months. In one instance, the fascia lata infolded between the whorls of wire and required a bronchoscopic dilatation of the lumen. Perhaps the whorls of wire were placed too far apart so that the infolding was permitted.

The difficulty of infolding of the fascia lata between separated loops of wire is obviated if one uses tantalum gauze. This provides a permanent skeletal structure, well tolerated by the body, easily managed and placed with no tension on the suture line. With its reinforcement by a strip of fascia lata, a good degree of air- and watertight seal can be obtained (although some investigators using it in animals were not so fortunate). The fascia lata is eventually absorbed and the granulation tissue can insinuate itself between the spaces in the mesh and provide a bed over which the rapid epithelization from the surrounding wound edges can spread. Rob and Bateman[6] have used this method to repair a large defect in the cervical trachea following excision of a thyroid cancer which had secondarily invaded it. After excision the aperture measured 9 cm. in length and practically involved the entire circumference of the trachea except for an 0.5 cm. area at its posterior margin. They lined the tantalum mesh gauze on both sides with fascia lata, and within a few days an inpocketing or "flutter" of the inner graft necessitated bronchoscopy and the touching of its surface with the electric cautery. As this was done, the inner graft was seen to shrink against the mesh and gave no further trouble. The lumen of the trachea has been observed endoscopically and has remained undiminished for at least one year.

The case to be reported is that of a malignant tumor of the trachea known to be radiation-resistant, and which had produced symptoms of repeated pulmonic infections and progressive impingement on the airway. The success of the repair of the trachea after the lesion was excised can be in large part attributed to the contributions made by many of the authors mentioned above and, in particular, to Rob and Bateman for their description of their use of tantalum mesh-fascia lata graft. The diligence of the nursing and resident staff, shown in this case, was a necessary adjunct to its successful conclusion.

REPORT OF CASE

The first admission of an 18-year-old white man to the Memorial Hospital was on Oct. 20, 1950.

History.—The family history was irrelevant. There was a history of polyneuritis in 1941, with residual bilateral foot drop, which was considered by a good pediatric hospital to be unrelated to anterior poliomyelitis. There has been no progression in any of his muscular symptoms since that episode. The history of his present illness dates from infancy, namely, at the age of 1½ years, when he had noisy breathing, which was thought to be caused by an "enlarged thymus." This area was treated by three or four roentgen-ray treatments, the factors of which are unknown, but no erythema of the overlying skin resulted. His symptoms disappeared following the radiation therapy. At the age of 3 years he had an attack of pneumonia which was described as severe.

He was well until the age of 15 years, when he had "pneumonia" characterized by rapid onset of fever and difficulty in exhalation, aggravated by exertion. Chest roentgenograms at this time were said to be negative. He was treated by antibiotics and was well after three weeks' bed rest. Seven months later he developed a second attack similar to the first, and required bed rest for five weeks. This attack was characterized by considerable weakness, and accentuation of difficult exhalation was again noted. Penicillin seemed to provide relief, and roentgenograms of his chest were again reported as negative. One year later, hemoptysis occurred for the first time, concurrent with a third bout of pneumonia. This subsided but returned again in five months, associated with all of the above-described symptoms.

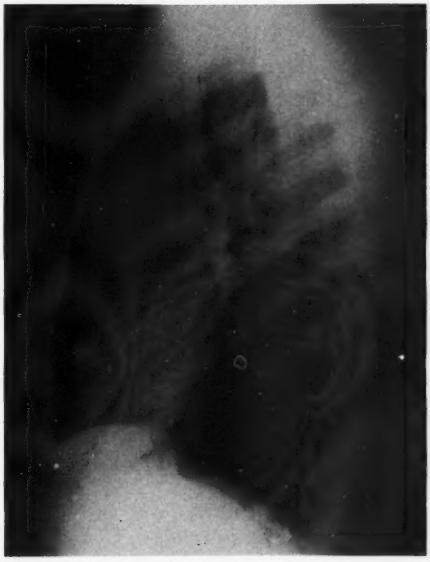


Fig. 1.—Lateral roentgenogram of chest showing oval-shaped tumor impinging on tracheal airway.

On bronchoscopic examination in July, 1950 (two and one-half years after the first attack of pneumonia as an adult), a tracheal tumor was found and a specimen for biopsy taken: this was reported as adenoid cystic carcinoma of the salivary gland type. Immediately following the biopsy, expiration and inspiration became easier, and remained so to the time of his admission to this hospital.

Physical Examination.—The patient was a tall, well-developed, 18-year-old white man who had acne vulgaris about the face and neck. He blushed and perspired easily and showed a marked tendency to dermatographia. The essential features of his examination were as follows: The trachea was in the midline. The chest showed equal expansion with normal resonance. The breath sounds were coarse but no râles were heard. The heart was not enlarged and the rate and rhythm were regular. The blood pressure in the left arm was 150/100, and in the right arm 170/100. A repeat recording on two occasions showed a constant level. Bilateral foot drop was present with only weak dorsiflexion of the right foot and absent dorsiflexion of the left foot. The left knee jerk was 3 plus, and the right knee jerk was 1 to 2 plus. No abnormal reflexes or changes in sensation were noted.

Laboratory Findings.—Complete blood count, urinalysis, and blood chemical analyses were essentially negative. The only abnormal finding was a prothrombin time of 28

seconds, the normal reading of that day being 15 to 20 seconds. The cerebrospinal fluid was found to contain 41.6 mg. of protein per 100 c.c., but no cells.

An electrocardiogram showed right axis deviation which was thought to signify hypertrophy of the right ventricle and/or vertical position of the heart.

Posteroanterior roentgenogram of the chest showed the lungs and osseous structures to be normal. There was no evidence of mediastinal widening, and the cardiac silhouette was within normal limits. However, on the lateral projection, in the midtracheal region there was a semicircular area of homogeneous density, the broad base of which was on the

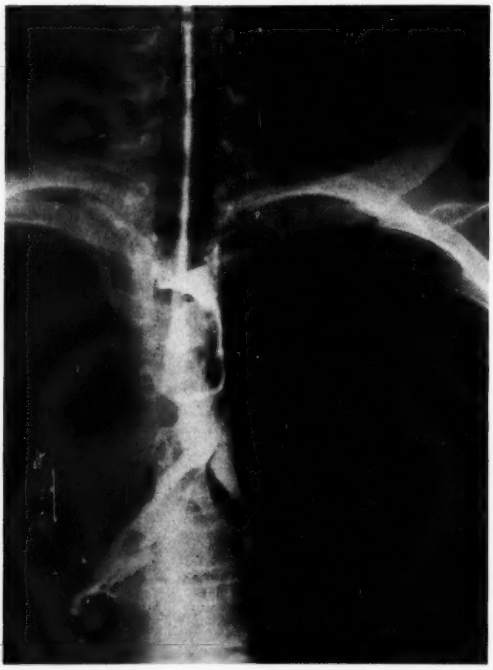


Fig. 2.—Tracheogram showing filling defect in trachea. Note its position above the bifurcation.

posterior wall of the trachea and the convex area projecting into the lumen (Fig. 1). The inferior margin of the tumor lay approximately 32 mm. above the carina, and a tracheogram showed that the tumor extended through the width of the trachea about 4 cm. in the supero-inferior diameter (Fig. 2). Lateral view of the tracheogram showed that the tumor protruded into the lumen approximately 2 cm., leaving a free airway about 5 mm. in width at its narrowest point. An esophagram was essentially negative.

Bronchoscopy on September 18 showed the superior border of a yellow, nonulcerating tumor, situated 24.5 cm. below the upper dental arch. The tumor projected into the tracheal lumen for at least 1 cm. and occupied the entire posterior wall, ascending for a short distance on the right lateral wall. It was estimated at this time that about one-half of the lumen was occluded, and the bronchoscope could not be passed beyond the tumor. Vital capacity and circulation time studies were normal.

Preoperative Preparation.—For six days preoperatively the patient was given penicillin aerosol (100,000 units of penicillin in 1.5 c.c. of zephiran aqueous solution), four times a

day. Beginning five days before operation, 300,000 units of crysticillin, twice daily; vitamin B complex; 500 mg. of vitamin C, daily; and 4.8 mg. of vitamin K, twice daily, were administered. On the day before operation, 1 Gm. of dihydrostreptomycin was started, and just prior to operation one more gram was given. Preoperative medication consisted of morphine sulfate, 1/6 grain, and scopolamine, 1/150 grain.

On October 26 anesthesia was induced with sodium pentothal and the cuff on the endotracheal tube was inflated with 4 c.c. of air. It was noticed that the airway was patent and the exchange good. Cyclopropane with 100 per cent oxygen was used throughout the procedure.

Procedure.—The patient was placed in the right posterolateral position and the chest and right thigh prepared with tincture of zephiran. The right pleural cavity was entered through the bed of the fifth rib after a small segment of the posterior portion of the fourth rib had also been excised. The tumor was found to lie just above the carina, on the posterior and right lateral surfaces, extending superiorly for about 3 to 3.5 centimeters.

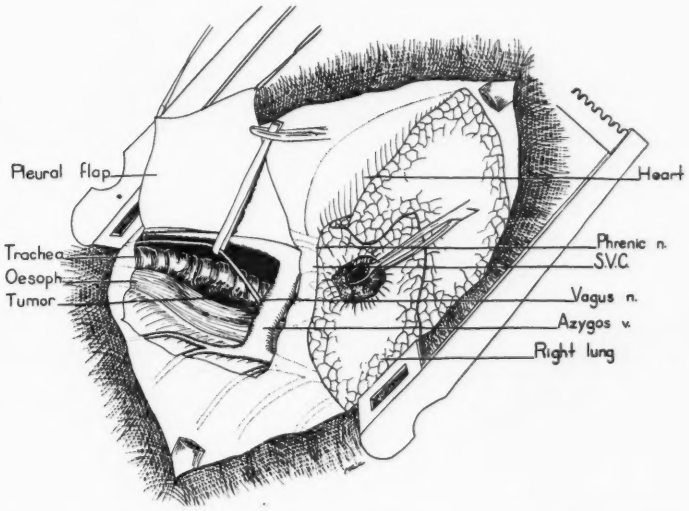


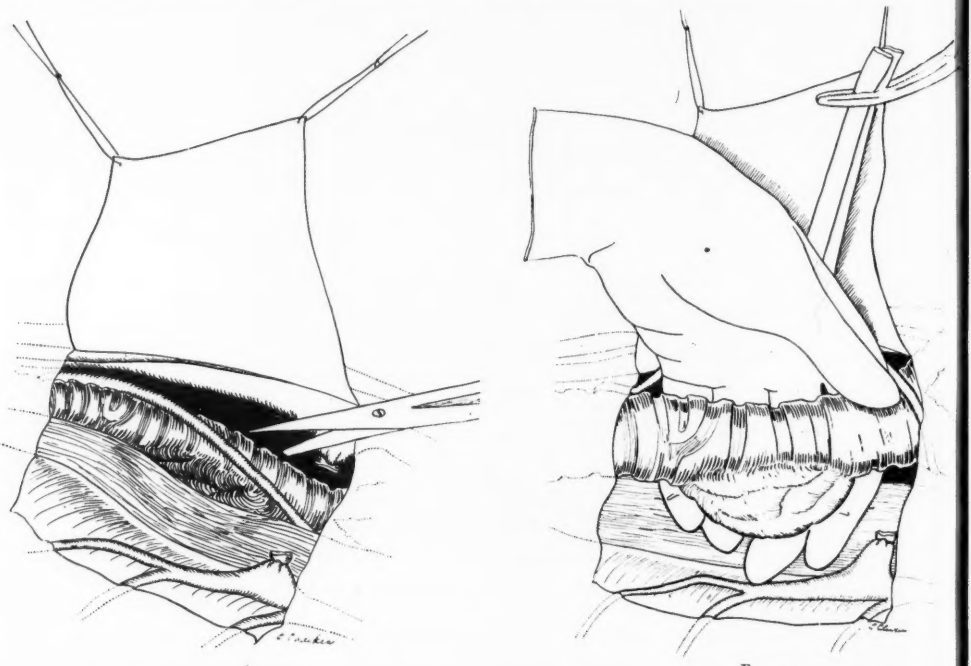
Fig. 3.—Over-all view of right thoracic cavity before azygos vein was divided.

It was fairly well defined, encapsulated, somewhat irregular and nodular, and indented the esophagus without being intimately adherent to it. No gross evidence of metastases was present. The tracheal tumor was palpable through the mediastinal pleura, and a pleural flap, considered to be of adequate dimensions to cover the area to be excised, was raised. The free edge of the flap was begun in the paravertebral gutter beyond the necks of the ribs; it measured about 12 cm. in width, with its base on the mediastinal pleura overlying the superior vena cava (Fig. 3). The right vagus nerve was freed from the tracheal wall and retracted, and the azygos vein next isolated, ligated and divided. The trachea was mobilized throughout its circumference (Fig. 4, *A*) from the superior portion of the mediastinum down to the carina (Fig. 4, *B*).

At this time an estimation was made of the dimensions of the tumor and the expected defect to be created. The tensor fascia lata was incised vertically and a strip of fascia, about 5 by 10 cm. in diameter, was removed; this was sutured to a piece of tantalum gauze slightly smaller than its over-all surface (Fig. 5). The tantalum gauze was infolded

about its periphery so that any sutures should not become weakened by possible fraying at its edges. A No. 000 plain catgut was passed through the fascia lata, then looped around the metal gauze, returned through the fascia and tied to the original suture. Multiple sutures of this type were placed about the edges of the mesh and at several points over its surface, so that there would be no shifting of the graft. For traction, a large Penrose drain was placed about the trachea, inferior to the tumor and just above the bifurcation of the bronchi.

An incision was made beginning in the cartilaginous rings, about 0.5 cm. beyond the palpable extent of tumor, and the lumen entered (Fig. 6). With the aid of a small hook retractor on the edge of the specimen (Fig. 7), the incision was developed, permitting a margin about the tumor of normal-appearing mucous membrane at least 3 to 4 mm. at its narrowest portion. As the posterior and left lateral portion of the tracheal wall was reached, the tumor-containing plaque was retracted posteriorly and the lumen widely opened.



A. *B.*

Fig. 4.—*A*, Beginning of tracheal mobilization by incision into areolar tissue between superior vena cava and trachea. *B*, Trachea and tumor isolated. Note division of azygos vein and retraction of vagus nerve.

The anesthesiologist, who had been maintaining respirations manually because the patient had stopped spontaneous breathing for some time, deflated the cuff, and a Kelly clamp was inserted into the orifice to pull down the lower portion of the endotracheal tube, pass it across the defect and into the left main bronchus (Fig. 8, *A*). This procedure was relatively easy, but when the anesthesiologist attempted to reinflate the cuff it was discovered that the clamp had made a rent in it. It thus became apparent that a closed system would have to be abandoned, and consequently the tumor was excised expeditiously. The resulting defect measured about 7 by 3 cm., and approximately one-half of the anterior portion of the tracheal cartilages was left intact.

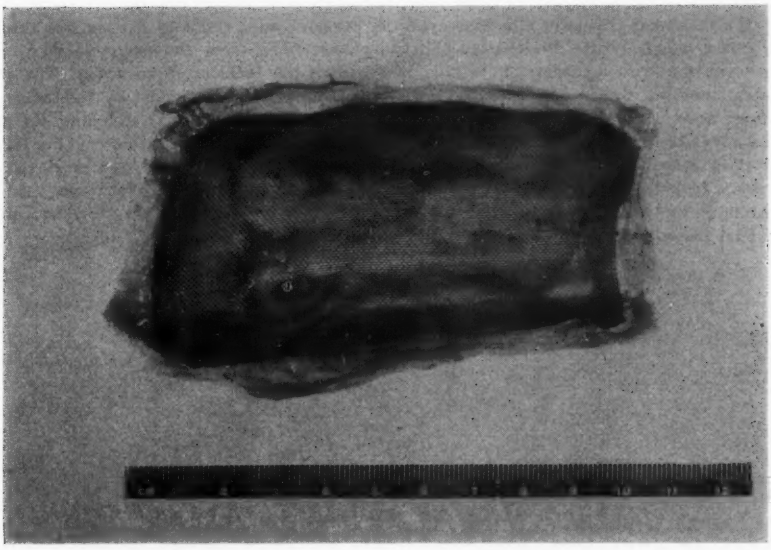


Fig. 5.—Tantalum mesh gauze and fascia lata sewn to it over multiple points. Note rolling of edge of mesh to prevent fraying and slipping of sutures at its margin.

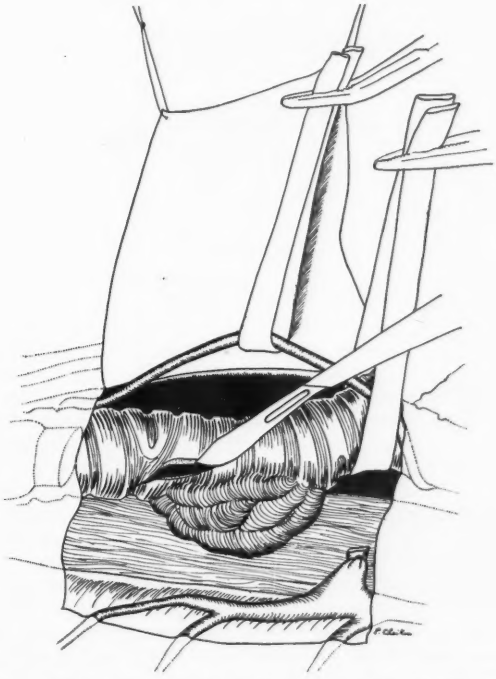


Fig. 6.—Incision into trachea. Penrose drain just above tracheal bifurcation. Right vagus nerve retracted.

At this point the patient became cyanotic, probably as a result of oxygen loss through the defect, coupled with diminished pulmonary excursions from the poor control of the closed system. Two maneuvers were used to overcome the problem of anoxemia: The endotracheal tube was drawn upward until its tip rested above the carina, and by tightening the Penrose drain that was in place above the carina about the trachea, occluding its walls sufficiently about the tube, a substitute for the broken balloon cuff was achieved (Fig. 8, *B*). The right lung, which was collapsed during the procedure, was inflated and employed for oxygen. In this manner the procedure progressed somewhat haltingly, for each time the right lung was collapsed to permit suturing, cyanosis would soon begin, so that sutures could be placed only in groups of four or five.

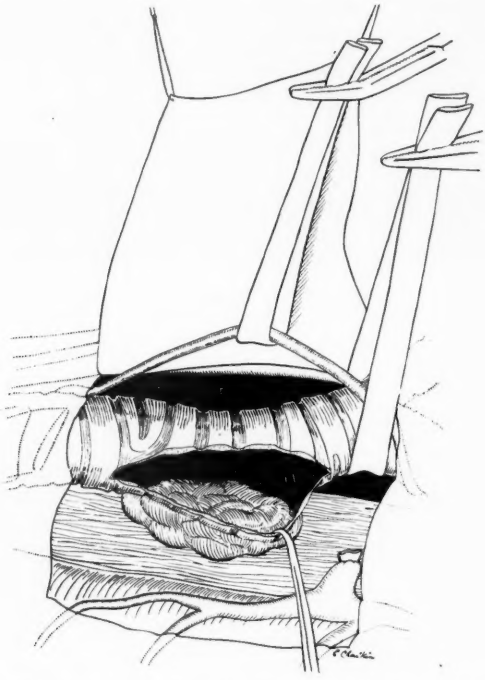


Fig. 7.—Retraction of tumor specimen to permit view of its interior extent.

The stent, composed of the fascia lata graft and the tantalum mesh, was held outside the thoracic wound in the position it would assume about the trachea, and a row of No. 000 plain catgut sutures was begun, uniting fascia lata and the superficial portions of the tracheal wall for a distance of 0.5 cm., beyond the edge of the left side of the defect in the trachea (Fig. 9). This was made feasible by rotation of the trachea throughout its long axis so that its opposite surface presented more readily. In addition, several No. 000 chromic catgut sutures were passed through the tantalum mesh and through corresponding portions on the tracheal wall about 3 mm. from its cut edge (Fig. 10). The graft was slid into position along these "guy ropes," and the sutures tied individually. The tracheal torsion was relaxed and the mesh and fascia lata were curved to cover the defect. The fascia lata was tacked to the tracheal wall by means of multiple silk sutures, in the form of a patch. As soon as this was completed, the trachea was relatively airtight, and the endotracheal tube could be used to better advantage for oxygen and anesthesia without

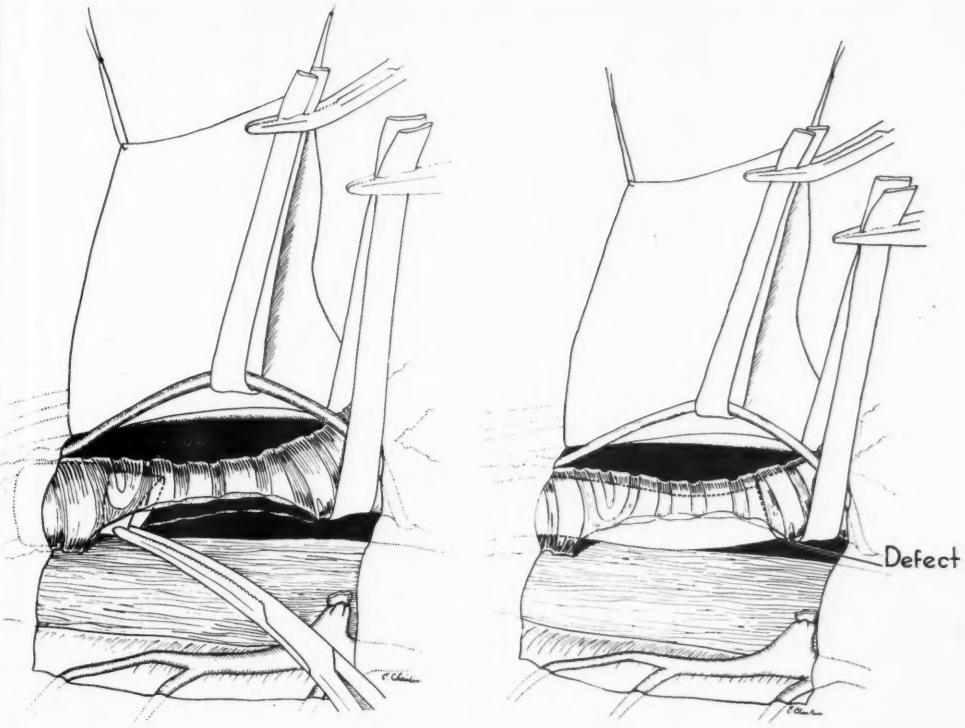


Fig. 8.—*A*, Clamp shown pulling distal tip of intratracheal tube with cuff deflated, down into the left main bronchus. *B*, Tube in place. Its position was maintained by the Penrose drain just above bifurcation because the balloon kept slipping out into the defect.

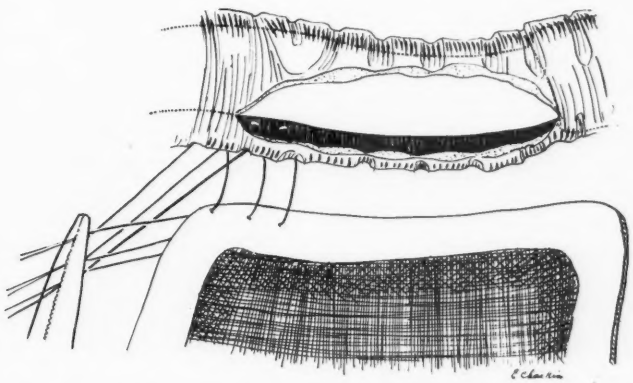


Fig. 9.—Beginning of posterior row of sutures through edge of fascia lata portion of stent.

the aid of the Penrose drain's circumtracheal compression. Retraction on the right vagus nerve was relaxed and the pleural flap that had been previously prepared was sutured over the graft with interrupted silk sutures, but not tacked too intimately to the inferior margin of the mediastinal defect so as to allow for possible egress of air (Fig. 11). A

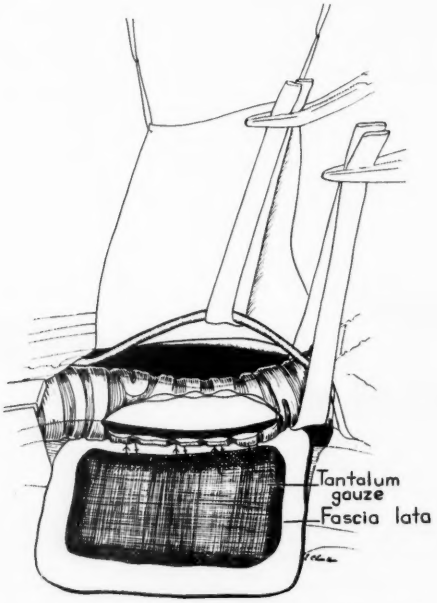


Fig. 10.—Interrupted sutures bringing tantalum mesh to edge of defect after posterior row of fascia lata sutures had been placed.

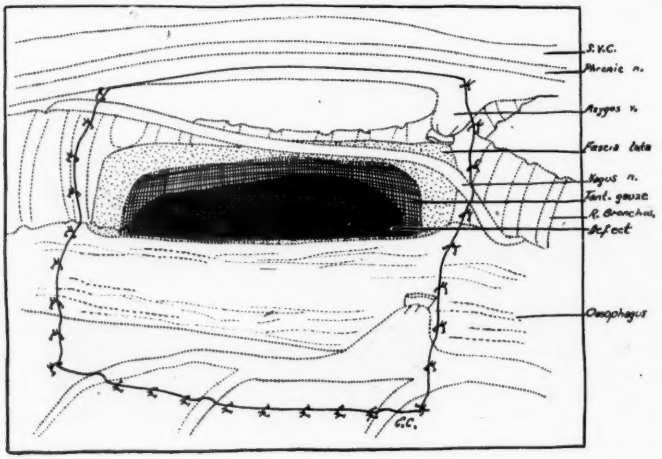


Fig. 11.—Completed repair demonstrating the pleural flap (in solid line); the fascia lata (stippled); the mesh (cross-hatched); and the defect (solid black).

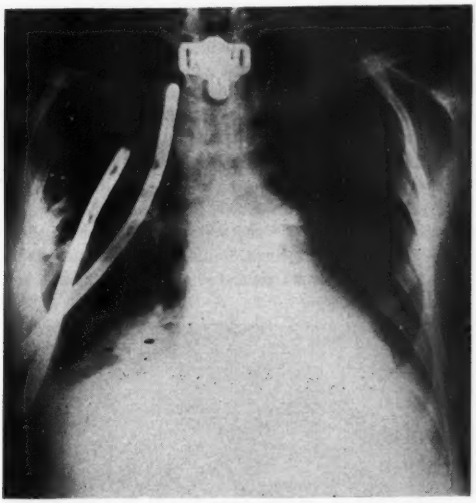
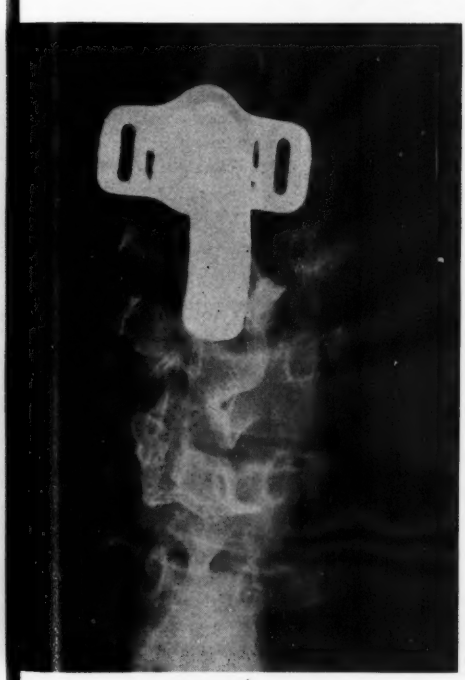


Fig. 12.—Roentgenogram of chest showing upper and lower underwater drainage tubes and tracheotomy tube in place (first postoperative day).



A.

B.

Fig. 13.—*A*, Front view of trachea showing tracheotomy tube and tantalum mesh. *B*, Lateral view of trachea showing tracheotomy tube and mesh. Note lack of diminution in size of tracheal lumen, and that upper margin of tracheal repair could be visualized through tracheostome when the tube was removed.

portion of the esophageal wall at the superior margin of the graft was sutured to the trachea to seal its upper end. Two hundred thousand units of penicillin, 1 Gm. of streptomycin, and 10 c.c. of saline were injected into the pleural cavity, and eucupine in oil was injected into the fourth, fifth, and sixth intercostal nerves. Two thoracotomy drainage tubes were inserted: one through the third intercostal space anteriorly and another below the thoracotomy wound in the eighth intercostal space. These were connected with individual underwater drainage bottles and the chest wall closed in layers.

The patient was then placed in the supine position and a tracheotomy performed with the insertion of a No. 8 tracheotomy tube. The purpose of this tracheotomy was to insure an avenue of suction, but in addition it was to decompress the tracheobronchial tree during exertion and coughing, resulting in less intraluminal air pressure referred to the suture line. His color was good and he was returned to the ward almost awake.

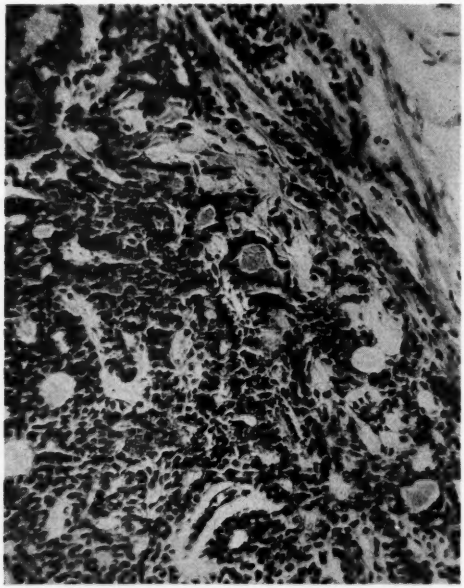


Fig. 14.—Photomicrograph of tumor called adenoid cystic adenocarcinoma, salivary gland type.

During the procedure he received 500 c.c. of normal saline and 1,000 c.c. of blood. His blood pressure at the onset of anesthesia was 130/70 (as compared with 165/105 obtained in his conscious state), and remained fairly static throughout.

Postoperative Course.—The patient was placed in an oxygen tent, and given 100,000 units of penicillin every three hours; 0.5 Gm. of dihydrostreptomycin every six hours; vitamins K, B, and C in preoperative quantities; and penicillin spray by De Vilbiss syringe through the tracheotomy tube. In addition, tracheal aspirations were done as needed. He ran a surprisingly uncomplicated postoperative course. Bubbling never occurred through the underwater drainage tube at any time, and both fluctuated for several days (Fig. 12). The suction material obtained from the tracheotomy was white and purulent for at least three postoperative days, and on the fourth day it began to clear. His temperature ranged from about 101° to 103° F. for the first postoperative week, gradually returning to normal on the sixteenth postoperative day.

Portable roentgenograms of the chest postoperatively showed the left lung to be entirely clear, and the right lung to have several densities (thought to be old lipiodol

deposits) scattered throughout the right central lung field. In addition, there appeared to be a small collection of fluid in the right pleural cavity, and on the fifth postoperative day this was tapped, yielding only 20 to 30 c.c. of clear fluid, and 300,000 units of penicillin were injected into the pleural cavity. Streptomycin was discontinued on his fourth postoperative day from which time 150 mg. of aureomycin, every six hours, was given. On the ninth postoperative day, the lower underwater drainage tube was removed; on the tenth day he was moving about the room, and on the twelfth day the second underwater drainage tube was removed.

On the twentieth postoperative day, the tracheotomy tube was removed to be cleaned, and it was possible to see the upper one-third of the defect in the tracheal wall (Fig. 13, *A* and *B*). At that time a sharp edge of the defect could be seen with no signs of inflammatory reaction about it. A yellow background was observed through the mesh, undoubtedly representing the fascia behind it. This excellent chance to observe healing of the graft in situ was utilized from that day until the twenty-third postoperative day when definite signs of "pinking" were observed through the mesh as the granulation tissue appeared. In addition, it was noted that epithelial ingrowths along the edge of the defect were beginning to decussate the mesh. On the thirtieth postoperative day, granulation tissue budding through the mesh was clearly evident, and the tracheotomy tube was removed permanently.

The patient was discharged from the hospital on the thirty-first postoperative day, and since that time has been relatively asymptomatic. Bronchoscopy, three months postoperatively, revealed no trace of any tantalum mesh and epithelization appeared to be about 100 per cent complete. Repeat bronchoscopic examinations were made every two months and showed that the lumen had maintained its diameter completely and that there was no evidence of tumor. His latest bronchoscopic examination on Oct. 31, 1951 (one year postoperatively), revealed no recurrence of the tumor or luminal narrowing. Four months later there was still no evidence of disease.

Pathologic Report.—The pathologic specimen measured over-all 3 by 3 by 2 cm. and showed an adenoid cystic adenocarcinoma, salivary gland type (Fig. 14). Dr. Fred Stewart states, "Of course, the behaviour of such a lesion in the trachea is *not* well documented, but it is extremely sluggish, and the extent to which this tumor can metastasize is virtually unknown."

SUMMARY

A case of carcinoma of the trachea, with excision and repair of the defect with tantalum mesh gauze and fascia lata, is reported. The patient is alive and without evidence of disease or change in tracheal lumen size sixteen months after operation.

A brief reference is made to various methods described in the literature which have been used to overcome the physiologic and reparative difficulties attendant upon such procedures, and their application to this case noted.

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LIPOTHYMOMA: AN UNUSUAL BENIGN TUMOR OF THE THYMUS GLAND

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BENIGN tumors of the thymus gland are extremely uncommon. Adenomas, fibromas, lipomas, and myxomas are said to comprise the majority of this group. Benign encapsulated mixed tumors composed of inextricably intermixed thymic and fatty elements are virtually unknown. As far as we are aware, an unequivocal example of such a neoplasm has been reported only once.[2] As in the case to be reported here, the patient was a child, and the tumor was not associated with myasthenia gravis or any other abnormality. Because of the unusual character and apparent rarity of this type of benign tumor of the thymus gland, the present case is reported in considerable detail.

CASE REPORT

P. O., a 10-year-old white girl, was admitted to the hospital during November, 1946, because of paroxysmal coughing of ten weeks' duration. She also complained of raising more sputum than normal and on two occasions the sputum had contained blood. Roentgenograms of the chest, taken one month prior to admission, showed atelectasis of the right middle lobe. The child had never suffered from dyspnea, although she became easily fatigued while playing. There was no clinical evidence of tuberculosis or of myasthenia gravis.

On admission her temperature was 99.6° F., pulse 80, and respirations 20. She was a slightly built girl, appearing younger than her stated age. A few anterior and posterior cervical lymph nodes were palpable. Anteriorly a region of increased dullness extended from the right of the sternum to the nipple line, and from the second to the fifth interspaces.

A chest roentgenogram revealed the presence of a mass in the anterior mediastinum. It was oval in shape, sharply outlined, rather dense, and extended laterally toward the right (Figs. 1 and 2). The leukocyte count was slightly elevated, being 10,400 per cubic millimeter. Nothing else of note was found.

Right anterior thoracotomy was performed three days after admission, and a well-encapsulated solid tumor was exposed in the anterior mediastinum. It was attached to the lower lobe of the thymus, and extended from the pericardium into the right thoracic cavity, compressing and rendering atelectatic the middle lobe of the right lung. The tumor was readily and completely removed.

Pathologic Report.—The specimen received in the laboratory consisted of a firm, encapsulated, oval tumor mass measuring 12.0 by 9.0 by 4.5 cm. in size, and, after fixation, weighing 170 Gm. The capsular surface was thin, smooth, glistening and transparent, but a few petechial hemorrhages, together with a few adherent fibrous tags, were present. On section, the surfaces exposed by cutting were firm, rubbery and pale yellow (Fig. 5). While the color was somewhat suggestive of a lipoma, the firm fibrous consistency of the tumor indicated that the mass was something more than encapsulated adipose tissue.

The entire specimen was fixed in 10 per cent formalin solution and blocks of tissue to be prepared for microscopic study were fixed both in 10 per cent formalin solution and in Regaud's solution. The sections were stained with hematoxylin and eosin.

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Received for publication Nov. 10, 1951.

†Dr. Ehler died on Nov. 5, 1951, after a brief illness.

Microscopic Description.—The tumor was found to be an unusual type of thymoma which was infiltrated throughout by normal-appearing adult adipose tissue and varying amounts of fibrous connective tissue. Sections taken from various portions of the growth, not only superficially but also deep within it, showed this admixture of thymus gland and fibro-adipose stroma to be uniformly present throughout the neoplasm, though the relative proportions of each varied in amount (Figs. 6 and 7). The thymic tissues were readily recognizable, being composed of accumulations of thymic lymphocytes in which were varying numbers of well-differentiated Hassall's corpuscles (Figs. 8, 9, and 10). The only variations from normal

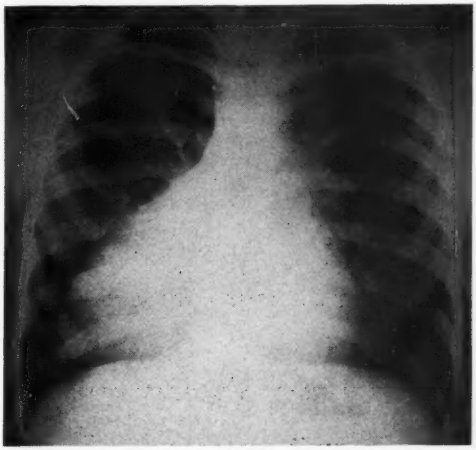

Fig. 1.

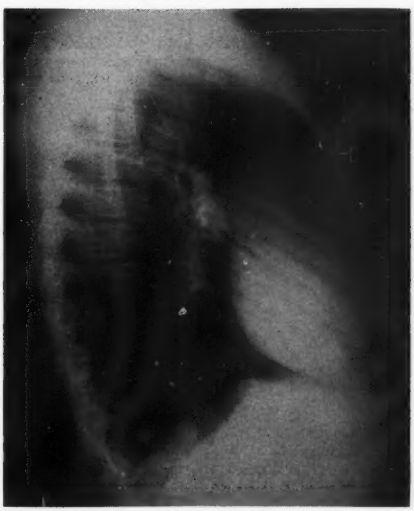

Fig. 2.

Figs. 1 and 2.—Roentgenograms taken before operation, showing the tumor as a large, well-outlined mass which appears to arise in the anterior mediastinum and to extend into the right pleural cavity.

thymic parenchyma were the distorted character of the lobules and the abundance of thymic tissue which appeared definitely increased in amounts. The lobules appeared either as isolated islands or as irregular narrow or broad anastomosing bands separated from each other by fatty stroma. There was also some loss of demarcation between cortical and medullary zones. While the stroma was composed chiefly of adipose tissue, it was mixed here and there with loose fibrous tissue. In some places the connective tissue actually predominated, being present either as bands of collagen similar to those which might be found in a fibroma,

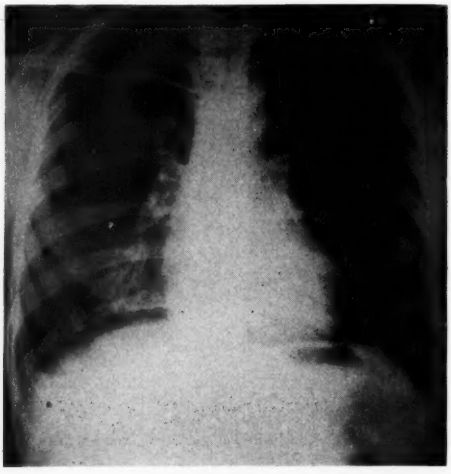

Fig. 3.—Roentgenogram taken after operation. The tumor is no longer present, and the thorax appears normal.

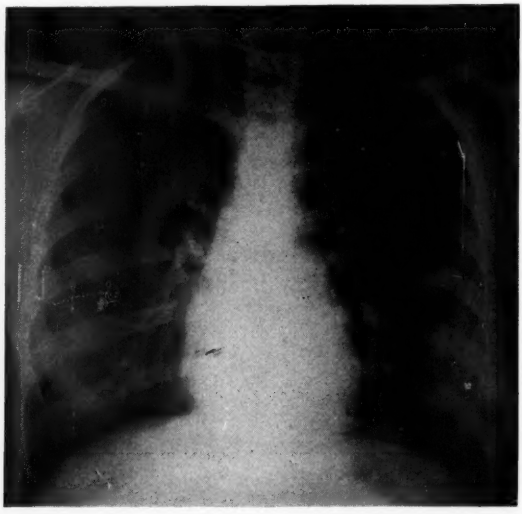

Fig. 4.—Roentgenogram taken four years after the tumor was removed. There is no change from the appearance of the thorax as shown in Fig. 3.

or as loose areolar tissue containing accumulations of fat cells within it. Sometimes the stroma was composed of adipose tissue alone. The most typical stroma was formed by both fibrous and adipose tissues with the fibrous tissue somewhat more predominant adjacent to the thymic lobules, while fat tissue was more abundant in the stroma further removed from the thymic elements. Thus, although the architectural pattern of the thymic tissue appeared altered and distorted by infiltrating fat and fibrous tissue, there was none of the disorderly tissue array that characterizes a malignant neoplasm.

The thymic tissues themselves appeared more abundant than normal suggesting that, as the neoplasm grew, thymic as well as adipose and fibrous tissues participated in the growth. The thymic tissue, however, became stretched or compressed as the neoplasm grew, with the result that it became a distorted, malformed structure within the tumor mass.

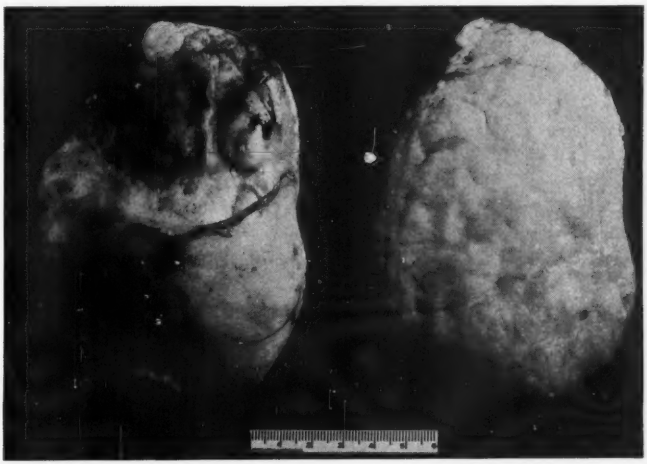

Fig. 5.—Photograph showing the capsular (left) and cut surfaces of the mediastinal tumor. The neoplasm is well encapsulated, solid, and homogeneous. Note large vessels in capsule and foci of subcapsular hemorrhage.

The over-all picture was that of a benign, well-encapsulated tumor composed of widely radiating strands of thymic tissue separated by proliferating fibro-adipose stroma. As indicated above, it appeared that both glandular and stromal elements participated in forming the benign neoplasm, thus making it a kind of mixed tumor, a fibro-lipo-thymoma or simply a lipothymoma, since adipose and thymic tissues predominated.

The child's postoperative course was entirely uneventful. Within four days the middle lobe of the right lung was completely re-expanded (Fig. 3) and she was discharged on the tenth hospital day. Periodic follow-up physical examinations during the next five years have shown no suggestion of recurrence of the tumor (Fig. 4), and there have been no signs or symptoms even suggestive of residual disability.

DISCUSSION

Since this girl has now lived for five years without any indication of recurrence, it seems reasonable to conclude that this unusual thymic tumor was entirely benign. Its large size, the symptoms it produced, and the roentgenographic findings presented no difficulty in establishing the presence of a mediastinal mass which needed to be removed. The operation was, therefore, completely successful.

Fig. 6.

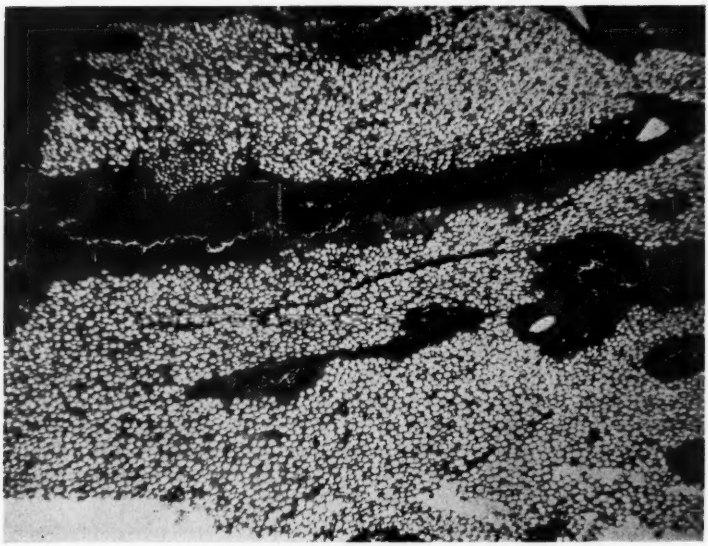

Fig. 7.

Figs. 6 and 7.—Low-power views showing the structure of the neoplasm. Note irregular, distorted anastomosing cords of thymus gland surrounded by narrow bands of collagen, and embedded in varying amounts of adipose tissue. (×30.)

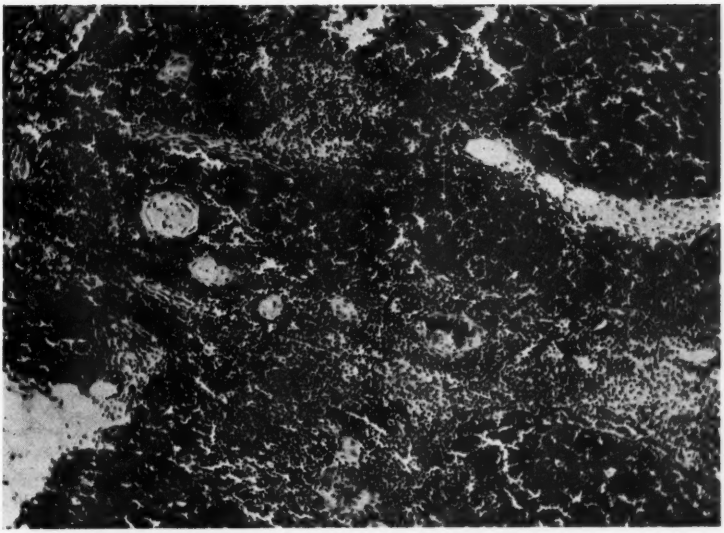

Fig. 8.

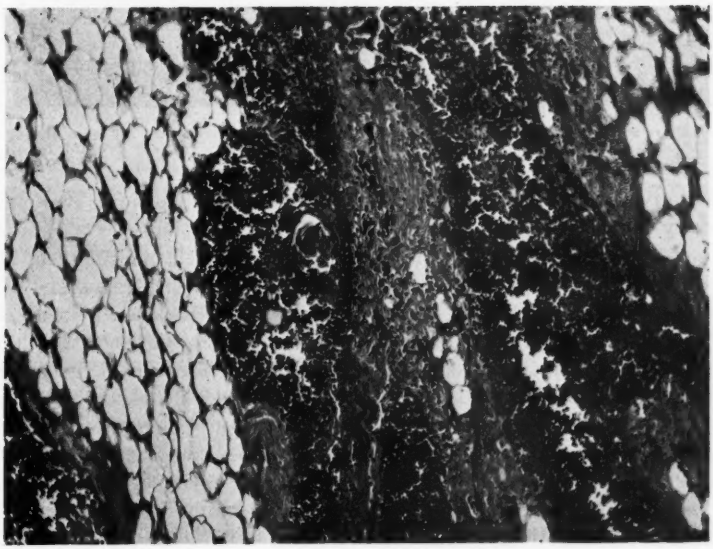

Fig. 9.

Figs. 8 and 9.—Sections showing typical thymic parenchyma embedded in adipose tissue. In Fig. 9 the presence of dense collagenous tissue adjacent to the thymus gland is clearly seen. Note typical Hassall's corpuscles. (×150, ×180.)

A review of the literature brings to light only one other unequivocal instance of an essentially similar tumor.[2] A few authors, in their descriptions of thymic tumors, suggest that they may have observed this type of thymic growth,[3, 14] but only the report of Andrus and Foot clearly describes pathologic changes that in most respects correspond to those observed by us. The size of the tumor in their case, 2,235 Gm., made it the largest thymic tumor ever recorded. In the years since their report no one else has encountered, or at least reported, any thymic tumor resembling this one.

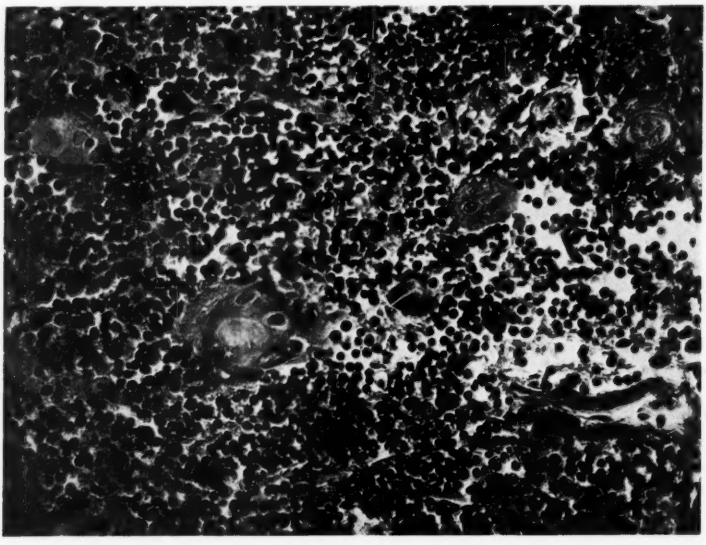

Fig. 10.—High-power view to show the typical thymic structure of the gland. Note thymic lymphocytes and reticulum and Hassall's corpuscles which were present in all parts of the tumor. (×550.)

Actually the changes of so-called involution of the thymus are characterized by atrophy of the gland with separation of the thymic lobules by a loose alveolar stroma. Descriptions of histologic sections from such glands, then, do not seem to differ significantly from the histologic features of this tumor. It has been noted also that with increasing age the adipose tissue component of the thymus increases proportionately, while the parenchyma on the other hand is decreasing.[13] Consequently, some question may be raised as to the nature of this large encapsulated growth. Is this mass really a neoplasm, or simply a form of fibro-adipose infiltration or an abnormal type of involution? The fact that the mass was circumscribed, localized, and of such large size makes it obviously a neoplasm, for it grew as an independent entity at a greater rate of speed than the normal tissues about it, ultimately forming the solid encapsulated tumor found at operation.

Is it a lipoma or a thymoma? This question cannot be so readily answered. In the strictest sense, a lipoma is an encapsulated mass of adipose tissue or fibro-adipose tissue which increases progressively in size, grows independently

of the surrounding tissues, and extends laterally by pushing adjacent structures to one side without infiltrating them. When various other components are also present, such as fibrous tissue, vascular tissue, etc., such terms as fibro-lipoma or angio-lipoma may be employed. When the adipose tissue appears less prominent than the fibrous or vascular elements, then the terms lipomatous fibroma or lipomatous angioma (lipo-angioma) may be employed. This tumor was a firm, rubbery, pale yellow neoplasm, ovoid in shape, encapsulated, and more like a fibromatous tumor than the soft, lobulated, bright yellow growth that is the typical lipoma. Therefore, this neoplasm did not seem to be a lipoma, although it did not resemble a thymoma either.

A thymoma, on the other hand, has always been difficult to characterize or classify. This is especially true of the malignant group where either lymphoid or epithelial elements, or both of them together, may participate in the neoplastic growth. With regard to benign thymic growths, the greatest difficulty is to differentiate between hyperplasia and neoplasia. Benign thymomas, however, characteristically possess a distinct capsule and histologically exhibit a distorted thymic structure often with a loss of cortico-medullary boundaries. Thickened trabeculae are generally encountered. Since most benign thymic tumors are adenomas, they are generally composed of solid masses of small round cells resembling lymphocytes infiltrated with thymic reticulum and Hassall's corpuscles. The observation has been made that fat characteristically does not infiltrate these tumors.[6] Consequently a benign thymic adenoma is an encapsulated, circumscribed tumor composed of structurally disarranged thymic elements often with dense dividing trabeculae, and usually devoid of adult fat cells.

The growth in our case, then, is characteristic of a benign thymic tumor which, however, is not a simple adenoma. For histologically the growth proved to be composed of thymic, adipose, and some fibrous tissues, thus making it a mixed tumor. The lymphoid elements, Hassall's corpuscles, thymic lobular arrangement, and thymic reticulum, unequivocally indicated its thymic nature. The noteworthy feature, as can be seen from the illustrations, is the marked and abundant admixture of thymic tissue with fibro-adipose stroma.

How shall a tumor of this type be named? Since the histologic nature and origin of many thymic tumors is still a subject of controversy, the organ designation with the suffix "oma" has served as the most satisfactory compromise for an admittedly unsatisfactory method of classification. This situation is not unique, however. For example, some tumors of the liver are still referred to as hepatomas; some which arise in the kidney are called hypernephromas; and those that arise in glial tissue are gliomas. It would seem appropriate, therefore, to designate this tumor a lipothymoma. If the expression, fibrolipoma, can mean a lipoma with fibrous tissue elements in it, then a lipothymoma may be interpreted as a thymoma with adipose tissue elements in it. The term thymolipoma might also be used, though thymolipoma emphasizes the lipomatous character of the growth rather than the associated thymic element. This unusual tumor, then, is considered to be a lipothymoma, for it is so obviously com-

posed of glandular and stromal tissues which grow as a true tumor. Moreover, both of these tissue elements appear to have participated in the formation of the neoplasm.

While many thymomas, both benign and malignant, may be associated with myasthenia gravis, in this case, and in that of Andrus and Foot, they were not. It is also noteworthy that both patients were children, one a 10-year-old girl, the other a 13-year-old boy. Both tumors were composed chiefly of adipose or of fibro-adipose tissue growing intimately and coordinately with thymic tissue. Although other unambiguous instances of similar neoplasms have not been recorded, or at least cannot be readily found in the literature, mention should be made of the report by Norris,[14] who recorded four instances of tumors associated with myasthenia gravis. His Case 1 was that of a 37-year-old woman with this disorder. At necropsy a thymic tumor weighing 57 Gm. with two cervical projections was found. The major mass of the tumor had a solid cellular structure, but sections from one of the projecting masses, designated as the left cervical neck, revealed a histologic picture similar to that of the tumor in our case. The tumor in his second case was in a 33-year-old white man who also had myasthenia gravis. This tumor weighed 17 Gm., and the histologic pattern was described as showing a lobular arrangement of thymic tissue with tonguelike projections separated by loose areolar tissue and dense fibrous tissue. Possibly both these tumors belong in this category of lipothymoma.

The thymic nodules reported by Alter and Osnato[1] do not resemble this entity, lipothymoma, as the title of their own paper clearly indicates although Andrus and Foot considered some similarity to exist. Furthermore, the tumor reported by Danisch and Nedelmann[8] is also in no way similar to this tumor type though it seems so regarded by Andrus and Foot. A translation of the title of their article clearly shows the dissimilarity. "A malignant thymoma in a 3½-year-old child with unusual metastases to the central nervous system." Perusal of this paper shows the histological pattern of their tumor to be clearly different.

It was thought that perhaps some of the recorded lipomas of the anterior mediastinum might be, in reality, tumors of this type.[5, 10, 11, 16] Possibly the thymic nature of yellow fatty-appearing tumors overlying the pericardium beneath the lower end of the sternum was simply overlooked. Beatson[3] records the case of a 45-year-old man in the anterior superior portion of whose mediastinum was an orange-sized lipoma. It was thought possible that this tumor might contain thymic elements. Unfortunately, no confirmatory microscopic studies were made. None of the other of more than fifty examples of mediastinal lipomas contains any reference to thymic tissue in fat. To be sure, histologic reports do not accompany all of the cases, yet, where recorded, mention is made only of mature fat, sometimes with an admixture of their connective tissue elements.

Finally, reference is occasionally made to lipomas of the thymus. However, no definitive bibliographic reference with histologic characterization could be located. For example Crotti[7] in cataloging tumors of the thymus states, "A lipoma has been reported by Wadde, Münchmeyer and Müller." Yet his bibli-

ography gives no reference which can be checked. Karsner[12] mentions that lipomas of the thymus have been reported. The only reference he cites in which lipomas are mentioned is that of Margolis.[15] Yet all this latter author says is that lipomas have been reported occasionally. His review and that of Bell[4] have recorded most of the thymic tumors in the older literature, but neither of them cites a reference to a report of a lipoma of the thymus gland. Furthermore Geschickter,[9] in an extensive review of lipomas, fails to record a lipoma of the thymus, although notations of such rare occurrences as a lipoma of the larynx and of most internal viscera is made. To be sure, an exhaustive search of the literature of the nineteenth century might turn up a concrete example. However, it is not certain that such a report would bear careful scrutiny. Was such a case from the mediastinum only? How was it determined that this lipoma was located in the thymus? Was it by gross observation alone? Was an involuted fatty thymus called a lipoma? Was an encapsulated mass of adipose tissue found in a thymus gland? It is even possible that this and other vague references to lipomas of the thymus refer to the entity herein described as lipothymoma, although we have been unable to verify this supposition.

Thus only one other unequivocal similar case is recorded in the literature[2] and two other plausible instances,[14] which were dissimilar in that they occurred in adults and were associated with myasthenia gravis. Of course tumors of the thymus are uncommon, and benign thymic tumors make up only a small percentage of this group. For that reason this entity, lipothymoma, will at most be encountered only infrequently.

FINAL SUMMARY

A rare benign thymic tumor composed of thymic and fibro-adipose tissues is described and its nature and possible origin are discussed. The neoplasm developed in the anterior mediastinum of a 10-year-old girl and was unassociated with myasthenia gravis. It is suggested that it be designated a lipothymoma.

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